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# TRAUMATIC AVULSION OF THE INSERTION OF FLEXOR DIGITORUM PROFUNDUS\*†

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IT is the intention of this paper to draw attention to a type of closed injury of the hand, hitherto undescribed, which is uncommon and perhaps of peculiar significance in the southern states of Australia, where Australian Rules football is played.

Each football season produces a few young men who have avulsed the flexor digitorum profundus tendon of the ring finger from its insertion. Because of its rarity there is a general unawareness of the entity, leading to delay in its diagnosis with a consequent effect on the success of treatment.

A description of the course of the condition, its clinical features, and its varied management is elaborated on the basis of eight cases which have been observed and treated in recent years.

#### AETIOLOGY

This injury occurs in healthy young males and is most often associated with participation in sport, notably Australian Rules football. The usual history given is that an attempt was made to hinder the man in front and the tip of the finger became hooked into his guernsey or shorts. As a result of the opposing forces a sudden pain was felt in the finger. This pain varies in severity and

may initially be disregarded during the excitement of the game.

It is of interest that in this series the ring finger was involved in every case. This is probably a reflection of the relative lack of independent action of this finger, demonstrated by clenching the first and then making an attempt to straighten each finger in turn while the other fingers remain actively flexed. The long flexors of the ring finger offer most resistance to this manoeuvre. A sudden pull on the ring finger tip while the clenched fist is firmly grasping an object provides the mechanism for this injury.

#### CLINICAL FEATURES

The typical clinical features of this condition undergo a progressive change with the passage of time after the injury.

#### Initial symptoms

The pain, swelling and stiffness apparent soon after the injury are usually explained as a "sprain" by the man, his team mates and his medical attendant. An X-ray examination is reported to show no fracture and the finger is often immobilized in plaster for a short period to relieve the discomfort and swelling. It is only later that the significance of the loss of active flexion at the distal interphalangeal joint is appreciated, often too late for optimum surgical treatment.

#### Early symptoms

During the first few days after the injury, the characteristic features are swelling of the finger, limitation of active movement at the

<sup>\*</sup>Based on part of a paper, "Injuries and Repair of Flexor Profundus Tendon within the Finger," read at the General Scientific Meeting, Section of Plastic Surgery, of the Royal Australasian College of Surgeons, Perth, 1958.

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proximal interphalangeal joint and absence of active flexion at the distal interphalangeal joint. The swelling and pain, often accompanied by inflammatory redness, may be severe enough to lead to a mistaken diagnosis of tenosynovitis. At this stage there is no flexion deformity of the proximal interphalangeal joint.

with, in addition, a flexion deformity of varying severity at the proximal interphalangeal joint in most cases (Fig. I).

The reason for the development of this flexion deformity is open to conjecture, but is probably associated with powerful traction through the intact vincula, so that the force

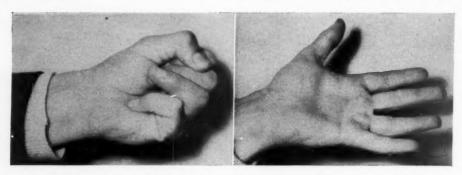


FIG. I. Typical clinical features in a boy of 17, eight weeks after injury (Case 4). Note restriction of active movement in both directions in the proximal interphalangeal joint, and the presence of a swelling in the proximal compartment of the ring finger denoting the site of the tendon end.

Careful examination will show that the tendon sheath over the middle phalanx is empty and it may be possible to feel the end of the profundus tendon. It may lie in one of three positions:

- (a) At the level of the proximal interphalangeal joint where it is held by unruptured vincula.
- (b) At the base of the finger, held by partially ruptured vincula. Here it may be free to pass backwards and forwards through the sublimis tendon and a painful clicking sensation is then experienced when the finger is
- (c) In the palm, after complete rupture of the vincula, where it produces a fusiform swelling in line with the affected finger.

Whatever the position of the tendon end, there is usually tenderness to deep pressure at the site.

#### Late symptoms

In those cases not seen for some weeks or months from the time of injury the inflammatory reaction will have subsided but the other findings are substantially the same, from both flexor tendons acts on the middle phalanx. This thesis is supported by the fact that the vincula remain intact in the majority of cases and all these cases develop a flexion deformity if neglected. In the only case seen several months after the injury that did not have this deformity, the tendon had retracted into the palm, a situation that can only occur if the vincula have been completely ruptured.

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Irrespective of the cause of the flexion deformity, if it is not relieved within a few weeks, secondary contraction will occur in the anterior capsule of the proximal interphalangeal joint. This change may be irreversible and may therefore prevent complete restoration of function even if the initiating factors have been corrected.

#### X-ray examination

An X-ray examination may show no obvious abnormality, but careful inspection of the film may reveal a small shadow lying in the anterior compartment of the finger close to the proximal interphalangeal joint. This represents a small flake of bone pulled off with the tendon and is a helpful confirmatory sign for those not familiar with the typical clinical picture (Fig. II).



FIG. II. (a) X-ray showing a small flake of bone lying in the anterior compartment of the finger at the level of the proximal interphalangeal joint (Case 7). This X-ray has been retouched; the shadows now clearly shown at the arrow point correspond exactly in size, shape and position to the shadows, too faint for adequate reproduction, in the original film.

(b) A much more obvious example, showing the flake of bone in the tendon end at the level of the neck of the proximal phalanx, together with bone chips scattered along the length of the tendon sheath, and a degree of subluxation at the diatal interphalangeal joint. It is significant that this case was referred within a few days of the injury.

#### TREATMENT

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There is now little doubt that optimum treatment, producing least delay, a minimum of technical difficulties and the best ultimate prognosis, is to replace the end of the tendon against the bare area at the base of the distal phalanx during the period before the tendon has irretrievably shortened, provided it can be found easily without prejudice to future action of the sublimis tendon. From experience, this period seems to be up to three weeks from the time of injury.

At operation, the flexor tendon sheath is exposed by raising a flap extending from beyond the distal interphalangeal joint to the base of the finger, as described by Rank and Wakefield (1953) for flexor tendon grafting. The flexor sheath is then opened just distal to the proximal interphalangeal joint, and the presence or absence of the end of the profundus tendon projecting through the bifurcation of the sublimis tendon is immediately apparent.

#### Re-insertion of the tendon (Group I)

In the ideal case with the end of the tendon presenting through the bifurcation of sublimis, the profundus tendon can be easily mobilized after dividing a few filmy adhesions between the "bare" tendon end and the surroundings. There may be no adhesions present.

The flexor sheath is resected from the level of the proximal interphalangeal joint to its distal end, except for a short segment, about three millimetres in length, at the midpoint of the middle phalanx. This segment acts as a pulley to prevent prolapse of the tendon during active flexion.

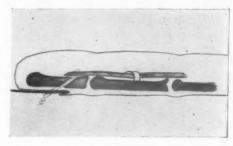


FIG. III. Schematic diagram showing a satisfactory method of attaching the tendon end to the distal phalanx.

The tendon is then brought down to the distal phalanx, after its end has been threaded through the pulley. This is a difficult manoeuvre owing to the snug fit, but can be facilitated by excising the expanded "wings" on the end of the tendon, without shortening it in any way.

A choice of procedures is available for reattaching the tendon to the terminal phalanx, or for attaching the distal end of a tendon graft should this be the operation of choice. weeks, still wearing the plaster, the patient attends regularly for supervised active exercises—no other type of physiotherapy is permitted. The strand of wire fixing the end

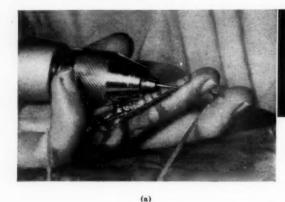




FIG. IV. (a) Shows the direction of the drill, with the point emerging through the nail.

(b) The wire twisted over the nail.

One method (Fig. III) is to drill two oblique holes through the phalanx. The drill point enters the bare area and passes backwards and distally to emerge through the nail distal to the nail fold (Fig. IV). A single strand of pliable wire is then threaded through the tendon as a simple loop and each end is passed through a drill hole; tightening the loop by twisting the ends over the nail (Fig. IVb) then holds the tendon firmly against the bare phalanx. This can be reinforced with a few simple sutures of silk between the end of the tendon and the surrounding soft tissues to produce a tidy

After four to six weeks, the loop of wire is very easily removed by cutting one end and pulling. No pull-out wires or buttons are needed and this method of fixation can be recommended for its efficiency and simplicity.

apposition.

An alternative method is to make use of the more secure Bunnell stitch; after insertion in the tendon the wire is threaded through drill holes in the terminal phalanx at right angles to the long axis. The stitch is then tied subcutaneously on the dorsum of the phalanx through a separate incision, and can remain in place permanently (Rank and Wakefield, 1953).

The whole hand is immobilized in plaster for three weeks. During the fourth and fifth

of the tendon is removed some time toward the end of this period. At the end of the fifth week the plaster is discarded and gentle use of the hand is encouraged. Suitable employment can be resumed at the end of the eighth week.

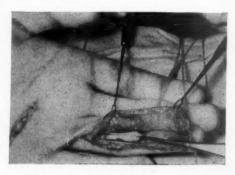


FIG. V. Same case as Fig. I (Case 4). Shows the tendon end lying free, but strong traction with forceps brings it as far distally as the proximal interphalangeal joint only. The tendon has shortened, and can only be brought close to the distal phalanx by acutely flexing the finger.

#### Tendon graft replacement (Group II)

In those cases where the tendon has shortened, and this may not be apparent until an attempt is made to re-insert it (Fig. V), or it has retracted through the sublimis tendon into the palm, there is a choice of two procedures. Either the useless profundus

tendon can be resected and replaced with a tendon graft (3 cases) or it can be resected and the terminal joint arthrodesed in a few degrees of flexion (3 cases). The choice between these procedures will depend largely on the extent of intrathecal adhesions; in the absence of adhesions tendon grafting is the method of choice even in the presence of a flexion deformity at the proximal interphalangeal joint.

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(a)

FIG. VI. (a) Shows a tendon graft prolapsing forwards in the absence of a "pulley" at the midpoint of the middle phalanx.



(b)

pulley over the middle phalanx is d sirable in the interests of ultimate efficiency of the graft (Fig. VI), but may not always be possible if the empty tendon sheath is collapsed and shrunken — yet another reason for early operation.

In most of the cases, correction of the tendon disorder, followed by active use of the hand, has resulted in spontaneous disappearance of the flexion deformity at the proximal interphalangeal joint. However, in one case (Case 3), it was found necessary to fit the patient with a gently acting traction splint, so designed that extension at the metacarpophalangeal joint was prevented and that active flexion of the finger could be produced while the splint was in place (Rank and Wakefield, 1953). Forceful manipulation permanently damages the joint, and must be completely avoided.

#### Arthrodesis of distal joint (Group III)

If it is thought that an attempt to restore active movement in the terminal joint is likely to prejudice the action of the undamaged sublimis tendon (see discussion), the efficiency of the finger can be improved by resecting the profundus tendon, and arthro-



(c)

(b) and (c) Shows a good range of active movement at the distal interphalangeal joint, but lacks the final "wind-up," i.e., relatively inefficient because the graft prolapses during flexion (Case 3).

The now standardized technique of tendon grafting is used, employing the tendon of palmaris longus for preference. Using one of the methods of fixation to the terminal phalanx already described, there has been no difficulty in obtaining firm union between the graft and the phalanx. Preservation of a

desing the terminal joint in a few degrees of flexion (Fig. IX). Of three cases where this was done (see Table), in one case the joint was excised and immobilized in plaster (Case 5), in another case the tendon had ruptured short of its insertion and a firm tenodesis was already provided (Case 6) and

in the third case the joint was excised and transfixed with a shortened Kirschner wire (Case 7) (Rank and Wakefield, 1953).

#### RESULTS

Typical cases grouped to illustrate choice of procedures available (see Table).

Group I — Re-insertion of the tendon (one case)

D.C., aged 17, schoolboy (Case 1). Injury to right ring finger at football nine days before operation (June, 1954). No flexion deformity at proximal interphalangeal joint.

tion (August, 1958). Had been X-rayed on day of injury — no abnormality seen, so nothing done. A few degrees of limitation of extension at the proximal interphalangeal joint (Fig. 1).

End of tendon was found emerging through the bifurcation of sublimis, no adhesions. Easily mobilized, but tendon had shortened and could not be brought down to distal phalanx without acutely flexing the finger (Fig. V). Profundus excised, free tendon graft of palmaris longus threaded through intact sublimis, with a short segment of the tendon sheath preserved as a sling at the midpoint of the middle phalanx. Fixed to phalanx with a single wire loop (Fig. III).

#### TABLE

Case	Age	Occupation	Aetiology	Finger	Time interval	Initial flexion deformity at proximal interphalangeal joint	Result
Group I-	Re-ins	sertion of the ter Schoolboy	dos. Football	Right	9 days	Nil	Full range active flexion at boti joints. Normal function (fou years) (Fig. VII).
Group II	-Repli 23	Student	ndon graft. Football	Left	10 days	Not stated	Full range active flexion both joints (six years)
3	15	Schoolboy	Games at school	Left ring	5 months	30°	Still some flexion deformity at proximal joint even after wearing traction splint for 6 months Graft prolapses (Fig. VI)
4	17	Schoolboy	Football	Left ring	8 weeks	A few degrees	Almost normal at both joints at four months (Fig. I, V, VIII)
Group III	Exc 22	ision of tendon,	arthrodesis of Football	distal joint Left ring	6 weeks	30°	Solid arthrodesis. Full range at proximal joint. Almost normal function (three years) (Fig. IX)
6	24	?	Football	Right ring	12 weeks	"Moderate"	Tendon had ruptured short of in- sertion and had produced a "natural" tenodesis. Profundus excised. Full range at proximal joint, four years later
7	19	Student	Football	Right ring	1 month	"Moderate"	Still had poor movement and flexion deformity at twelve weeks. Twelve months later, same result as Case 5
Group IV-	-No to	Farmer	Football	Left ring	3 months	Nil	Tendon retracted into palm. No flexion deformity

End of tendon emerging through bifurcation of sublimis, not adherent. Tendon not shortened, easily mobilized. Fixed to phalanx with a single wire loop.

Complete immobilization for three weeks, wire removed during fifth week.

Reviewed July, 1958. Full range of active independent flexion at both interphalangeal joints. Slight limitation of complete extension at the distal joint, but none at the proximal joint. No functional disability (Fig. VII).

Group II — Resection of profundus and tendon graft replacement (three cases)

R.M., aged 17, schoolboy (Case 4). Injury to left ring finger at football eight weeks before operaComplete immobilization for three weeks. Wire removed at end of six weeks.

Reviewed December, 1958 (four months after operation). No flexion deformity. Full range of active movement at proximal interphalangeal joint, and a good range at the distal joint (Fig. VIII). No functional disability.

Group III — Resection of profundus and arthrodesis of the terminal joint (three cases)

M.J.S., aged 22 (Case 5). Injury to left ring finger at football six weeks before operation (September, 1955). Limitation of extension at the proximal interphalangeal joint by about 30°.



(a)

FIG. VII. End result after four years from re-insertion of the tendon end in a boy of 17. Almost complete "wind-up" of the finger. Very slight limitation of extension at the distal interphalangeal joint, but no disability (Case 1).

End of tendon was at base of finger, with some adhesions and shortening. Profundus excised, and distal interphalangeal joint arthrodesed by excising the joint surfaces.

Reviewed July, 1958. Solid arthrodesis of the distal interphalangeal joint, with full range of active movement in the proximal joint. Very little functional disability (Fig. IX).

#### Group IV - No treatment (one case)

J.K.P., aged 30, farmer (Case 8). Injury five months previously to right ring finger while playing football. Only complaint was of a tender fusiform swelling in the palm in line with the finger. The absence of active flexion at the distal interphalangeal joint did not cause him any disability and he declined operation to remove the tender profundus tendon from the palm.

#### DISCUSSION

Early diagnosis is the key to success in the treatment of this condition. The main cause of delay in diagnosis seems to be a lack of

awareness of the condition; the production of an X-ray film that shows no fracture in the finger after a football injury adds to this difficulty.

If the diagnosis is made early enough the simpler procedure of re-apposing the avulsed tendon to the bare area at the base of the distal phalanx can be performed without the necessity for either a tendon graft or an arthrodesis of the terminal interphalangeal joint.

The best result in this series, and the one obtained by the simplest operative procedure was that in Case 1 (Fig. VII), where the tendon was replaced directly to the terminal phalanx. This procedure carries the least risk of producing intrathecal adhesions, provided the tendon sheath is not opened proximal to the proximal interphalangeal joint.





FIG. VIII. Same case as Fig. I. End result after four months from replacement of avulsed tendon by a free tendon graft of palmaris longus threaded through the intact sublimis tendon. Still lacks the final "wind up," but a good range of active flexion at the distal joint. No limitation of extension at the proximal interphalangeal joint, and very little at the distal (Case 4).

However, it must be admitted that several of the other cases show a result not far short of this, whether a tendon graft replacement was performed, as in Case 2 and Case 4, or whether the terminal joint was arthrodesed after excising the profundus tendon as in Case 5. If these cases are examined critically, minor anatomical deficiencies are apparent, but many of these patients claim to have no functional disability for most activities.

Whether movement is re-established by replacing the profundus to the terminal

familiarity of the surgeon with the finer points in the technique of tendon grafting.

Although the results of tendon repair or graft are inclined to be disheartening in the early post-operative period, the long term results are most encouraging. These men are almost always young, healthy types of above average intelligence — good subjects for reparative surgery where the enthusiasm of the patient is almost as important as the quality of the surgery in obtaining the final result.





(a)

(b)

FIG. IX. End result after three years from resection of profundus tendon and arthrodesis of terminal joint. Note the full range of movement at the proximal interphalangeal joint. Good functional result. (Case 5).

phalanx, or by tendon graft replacement, preservation of a small segment of the flexor sheath at the midpoint of the middle phalanx is essential if the tendon is not to prolapse during flexion and thus become relatively inefficient, as in Case 3 (Fig. VI).

Tendon graft replacement undoubtedly carries the greatest risk of producing intrathecal adhesions, so that instead of a finger whose sublimis tendon is acting over a reasonably good range, one may end up with a finger with little or no active movement at either interphalangeal joint. Therefore, if there is any possibility that threading a tendon graft through the bifurcation of sublimis will make matters worse, it is far better to accept resection of the profundus and arthrodesis of the terminal joint as the optimum As has been shown, this proprocedure. cedure produces a most useful finger with only slight functional limitations. which may influence this choice are the presence of any but minimal intrathecal adhesions at the time of operation, and un-

#### SUMMARY

- Traumatic avulsion of the insertion of the tendon of flexor digitorum profundus of the finger is described.
- The probable mechanism responsible for the injury and the subsequent deformity in untreated cases is discussed.
- Various procedures undertaken at different stages in the progress of the injury, and under varying conditions, are described and discussed.
- The long term results in eight cases are demonstrated.

#### ACKNOWLEDGEMENTS

My thanks are due to Mr. J. T. Hueston for permission to publish Fig. IIb, and to Mr. B. K. Rank and Mr. A. R. Wakefield for allowing me to include cases treated by them in this series.

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#### PSYCHOLOGICAL ASPECTS OF SURGICAL PRACTICE\*†

By DAVID C. MADDISON

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S OMEONE said recently that, although the status of the psychiatrist is improving in the eyes of his professional colleagues, he still goes into dinner at the rear of the medical field, a few paces behind the gynaecologist. The background of this low reputation could well become a paper in itself; my purpose here is to attempt to show that the specialist in psychological medicine has a great deal of theoretical and practical value to offer his associates, a contribution which is available less frequently than it should be because of the still prevalent notion that psychiatrists have a useful function only in the treatment of gross madness. The surgeon, as much or more than any other doctor, is in all his transactions with patients dealing with entire human beings as psychosocial units, individuals with minds and bodies interacting with their current and past environments, and possessing diseased or damaged organs as one aspect only of their total behaviour. Increasing specialization is the most serious disease of medical practice in our age, and "the patient-as-person has been threatened with extinction by fragmentation, leaving behind him an odd assortment of calibrated readings, metaplastic cells, radioactive isotopes, and several feet of glass tubing, which, assembled in whatever combinations imaginable, fail to add up to a recognisable facsimile of man" (Meyer, 1958). To those surgeons in particular who still live under the delusion that skill in their specialty can be equated purely with technical proficiency and who are wholly or partly unaware of the significant repercussions which both major and minor illnesses will inevitably have on the psyche of their patients, this paper is particularly directed.

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#### BASIC PSYCHOLOGICAL CONCEPTS

It is often complained that psychiatrists are largely or entirely incomprehensible to their colleagues because of the exotic language in

which their statements and reports are expressed. Despite the kernel of truth in this accusation, it more correctly represents the position to point out that the language of the psychiatrist is virtually foreign to his colleagues because of the woeful deficiency which exists in psychiatric training in contemporary British medical student curricula. This failure of communication is nowhere more apparent than when the concept of "unconscious" motivation is introduced, a concept which lies at the core of modern psychological theory yet which is totally unfamiliar to the majority of non-psychiatrists; to the surgeon, motivation and feeling which is not readily apparent and objectively demonstrable tends to be regarded as non-existent, whereas the psychiatrist spends a large part of his professional life in the repeated search for unconscious meaning and purpose in the thoughts, actions, character and attitudes of his patients. The basic considerations which follow are liable to be thought incomprehensible and artificial if this is not borne in mind; what may appear of dubious validity when baldly expressed in print can be repeatedly shown to be of vital importance when the surface layers of the personality are made less significant by drugs, by hypnosis, or in psychological treatment.

#### 1. The development of the body image

Observational studies of infants show that initially the very young child is unable to distinguish between himself and the outside world; by gradual motor exploration and by exposure to a variety of sensory stimuli he comes to develop a relatively clear differentiation between that which is his own body and that which is not. Children's drawings show that the mental picture a young child has of his own body is far from accurate, but with further growth and the experiencing of new internal and external sensations, there develops what is known as the body-image, "the picture of our own body which we form in our mind . . . the way in which the body appears to ourselves" (Schilder, 1950). This is more than a simple summation of sensations; mental pictures are involved additionally, and combine to form an image which

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<sup>†</sup>Based on a paper delivered at a seminar in the Department of Surgery, University of Sydney, on 8th July, 1958.

tends to remain outside central consciousness. Though few of us are so introspective as to bring this schema more than occasionally into conscious awareness, a little reflection should convince us, with Schilder, that "the body-image is one of the basic experiences in everybody's life . . . one of the fundamental points of life experience".

The integrity of the body-image is an important determinant of an individual's ability to adapt to external reality in a healthy and confident fashion, and any circumstances which impair, or threaten to impair, this integrity will inevitably be productive of some degree of anxiety, whether this is experienced consciously or not.

The body image is not by any means identical with the objective limits of the body in reality. The most obvious example of this proposition is seen in the phenomenon of the phantom limb or breast following amputation, when a lengthy period of time may elapse before the body-image and the objective body again co-exist. Those parts of the body which come into a close and varied contact with external reality, i.e. the foot and hand, are those which tend to persist for the longest time in phantom form, and peripheral explanations of such findings must clearly be inadequate. Sudden loss seems to be a prerequisite for phantom development; an individual's long held perception of himself as a complete person cannot be altered overnight, and the old familiar pattern is retained or reactivated by emotional forces attempting to resist at a psychological level the experience of mutilation. The influence of higher emotional processes on this state can be illustrated by the capacity of a shortened phantom to be lengthened during and after the induction of an hypnotic trance.

In addition to the striking phenomenon of phantom formation, other less obvious ways exist in which the body image can be shown to extend beyond the confines of the material body. Our clothes become invested, even in the normally adjusted individual, with a certain modicum of mental energy and interest, which may become pathologically exaggerated in certain neurotic states; it is certainly a universal experience that a change of clothes may radically alter our mental set and perception of ourselves for a brief period. The behaviour of certain car owners, either

through the aggressive manner in which their vehicles are used to express their own conflicts, or by reason of the scrupulously obsessional care lavished on their upkeep, strongly suggests that the image of the body may be extended to embrace even an automobile; the practice of certain American adolescent gangs of attaching two coconuts to the rear axle of their "hot rods" is of interest in this connection! Recognition of movement, locality and distance may extend well beyond the body limits to the end of some familiar instrument held in the hand. and every motorist recognizes the difficulty in judging distance and width when driving a new vehicle; the instrument, or the older car, has become part of our body image.

Disturbances of the body image are of common occurrence in psychiatric practice, but are only very infrequently of primary importance to the surgeon; there are, however, rare instances in which he may find himself confronted by a patient whose somatic complaints mark a disruption of the psychic unity of the body.

#### Case 1

A young woman of 23 years complained of difficulty in swallowing for which she had been medically investigated before passing into psychiatric care. More detailed history-taking revealed that she believed her food, after passing her throat, deviated to the right of the midline and ended up in the bottom of her right breast, These complaints were one feature of a schizophrenic disorder.

#### 2. Narcissism

Dynamic psychology acknowledges the existence of only a fixed amount of mental energy, the neurophysiological substratum of which is, of course, unknown at present. The normal individual uses a substantial part of this to form satisfying relationships with other people, objects and activities, and the ability to do this is determined in large part by the gratifying nature of the relationships which the young child forms with the first individuals in his experience, his parents or parent-substitutes. Failure to establish such comforting early relationships may lead to what the individual unconsciously conceives to be a safer disposition for his psychic energy, as a result of which he turns a disproportionately large amount towards his own body and its functions: here, at least, there is felt to be no threat. When this selfinterest is excessive, the resultant attitude of

the person is described as 'narcissistic'. Such a mechanism exists in any form of hypochondriasis, and is probably not essentially different from the extreme self-concern of some men and many women who show excessive preoccupation with their bodily health, fitness or beauty.

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But these are pathological exaggerations of the normal degree of interest which all mentally healthy people take in the appearance and welfare of their own bodies. Such interest is inevitably increased in physical illness, and to some extent every sick person tends to become narcissistic; much of his mental attention is focussed on his diseased organs, and his other interests and relationships tend to be relatively deprived of genuine emotional participation. Most especially is this so when illness or injury concerns the head or the genitals, for these two areas are those most intimately connected with the individual's conception of himself as an intact and fully differentiated human being - in other words, these areas are those most intimately connected with the body-image, towards which even normally a substantial amount of mental energy is directed. Considerations of this kind are doubtless responsible for the finding that serious depressive reactions are more frequent in female patients who have experienced pelvic surgery than following cholecystectomy (Lindeman, 1941); surgical attack in the genital area is seen as a greater threat to individual integrity than operations elsewhere.

#### 3. Castration anxiety

Both observational studies of young children and material derived from retrospective psychological investigation of adults show that there is a regular and consistent reaction in childhood to the perception of the anatomical difference between the sexes. The fear of punishment by castration in boys may be reinforced by angry or jocular threats from adults to "cut it off", usually uttered as a response to the discovery of infantile masturbation, which in any event is ubiquitous; young girls may believe themselves to have been previously punished in this way for a similar offence. The conflicts arising from this period, referred to loosely as "castration anxiety", are dealt with by the psychological mechanism of repression, and thus are not accessible to conscious awareness in adult life; nevertheless they may play a substantial

part in much later neurotic behaviour, and this anxiety (or an attempt to deny its existence) makes understandable many otherwise odd and perplexing facets of human behaviour, including, for example, some obscene jokes which are based on a thinly disguised castration theme. More importantly in our present context, repressed anxieties of this nature play a large part in determining many patients' exaggerated fears of doctors, and most particularly their anxiety concerning surgical procedures. Even when the operation is not specifically directed at the genital area it may be unconsciously perceived by the patient as symbolically equivalent to castration, especially if the surgical assault is of a mutilating kind, and may reactivate all the old anxieties and fears. When the operation is aimed at the sexual organs, this factor will be substantially reinforced.

#### Case 2

A man of 53 without previous history of psychiatric illness was submitted to a circumcision operation about which he felt considerable apprehension. He specifically and voluntarily stressed when interviewed subsequently that he was most disturbed in the immediate post-operative period because, in the small private hospital in which the operation was carried out, he was asked to use the female toilet as the male toilet was temporarily out of action. Two weeks following surgery he developed the first signs of a severe psychotic depression.

In the older child, also, circumcision may have a considerable emotional investment, particularly if the child experiences the operation at a period when he is exposed to concurrent psychological stresses.

#### Case 3

An extremely neurotic young man receiving analytic psychotherapy was puzzled by the occurrence of a vivid anxiety dream in which he was standing beside a tall wireless mast which was gradually toppling to the ground. With the passage of time in treatment he was able to recall that the first dream of this nature had taken place while under anaesthetic for circumcision at the age of 7. He was an illegitimate child with an extremely disturbed relationship to his mother, and who felt considerable guilt concerning early masturbation and sexual play; one of his major neurotic symptoms in adult life was severe impotence. The meaning of the dream was clearly explainable in terms of orthodox psychoanalytic symbolism.

These basic psychological concepts have been introduced and described in an attempt to throw more light on the serious threat which surgical procedures represent to the child, adolescent, or adult patient, whether he manifests a "normal" or neurotic character structure. In every instance the body-image of the individual is to some extent threatened by the possibility of surgical assault, and this threat is heightened if the operation is a mutilating one and if it is directed towards the genital area, as in both instances narcissistic and castration anxieties may be unconsciously evoked. In the patient of neurotic character, even if he lacks a past history of overt neurotic symptoms, the possibility of psychic disturbance is correspondingly greater by reason of the more intense degree of narcissistic orientation and of residual castration fear in his mental make-up.

#### SPECIFIC CLINICAL PROBLEMS

The areas of mutual significance for psychological medicine and surgery are numerous and interlocking, and the one patient may show psychological reactions of interest and importance at several points from the time of his development of illness to the stage of post-operative convalescence. The following review is thus divided into what are essentially artificial subdivisions, and is intended merely to highlight certain relevant clinical situations, in all of which our knowledge is small and (with significant exceptions) largely anecdotal, and where the opportunities and need for further research are real and urgent.

#### 1. Delay in seeking treatment

Little is known in any systematic fashion concerning the relevant factors involved in causing sick people on all too many occasions to permit unnecessary delay to intervene before seeking medical attention for serious complaints. This problem has relevance for many areas of surgical practice, but nowhere more than in the field of malignant disease; the work of organisations devoted to public education in cancer detection is seriously impeded by this factor, as procrastination and postponement of medical consultation is not by any means confined to those who are of low intelligence or ignorant as to the nature of malignancy. Much importance must be attached to the psychological mechanism known as "denial", that is, a mental set found in some neurotic adults which permits them to indulge, like a child, in wish fulfilling fantasies even in the face of reality factors which show their beliefs to be palpably false.

Case 4

A woman of 65, of high intelligence and with several medically qualified relatives, detected a hard lump in her breast eight months before she sought medical advice; the condition was of malignant nature and surgery and radiotherapy were unable to control its spread. Even superficial psychological study showed an intensely neurotic pattern of behaviour throughout her life, with a constant tendency to retreat from recognition of unpleasant realities rather than face them and deal with them in a constructive fashion.

Aitken-Swan and Easson (1959) found that 19 per cent. of 231 patients who were directly informed by consultants of the existence of cancer were still using the denial mechanism up to one month later; this reaction was more frequent in men than in women. What other factors are concerned in behaviour of this type are as yet unknown, but further studies in this field are being carried out at McGill University (Henderson et alii, 1958).

#### 2. Pre-operative anxiety

It is a matter of common observation that patients vary greatly in the degree and quality of the pre-operative anxiety which they manifest, and numerous factors assist in determining this difference. The extent to which a patient's psychological state may vary from the pre-operative to the post-operative period has been neatly shown by Meyer et alii (1955), who administered a psychological test to a group of patients both before and after surgery and noticed a striking difference in the results on the two occasions; the test used is one in which the patient is asked to draw a house, a tree and a person, from the interpretation and analysis of which an experienced clinical psychologist may come to significant conclusions as to the person's current emotional state. In the normal individual these drawings show a remarkable constancy over a period of time, but in the surgical group the pre-operative test showed many indications of intense anxiety, which was being dealt with by the mechanism of regression, that is, a retreat to a less mature, more childlike form of behaviour; these signs subsided substantially in the post-operative period.

It is not appropriate to assume that preoperative anxiety will of necessity be to the patient's detriment. Titchener et alii (1957), in one of their several papers describing psychological observations on 200 randomly selected surgical patients, found that increased anxiety and/or fear appearing pre-operatively

were associated with a good or improved peradjustment in the convalescent period. Stengel et alii (1958) found that intense pre-operative emotional reactions were only very exceptionally followed by postoperative mental disorder. Certainly it is desirable that such anxiety should be able to be freely ventilated; many patients will attempt to deal with their apprehension at this time by the unhealthy mechanism of denial, and in this immature and potentially harmful behaviour they are all too often encouraged by the surgeon and by nursing staff who may find themselves too busy to encourage the patient to talk about his fear so that it may be adequately understood and put into perspective, and instead may deal with the situation by glib reassurance and evasions which only increase the patient's uncertainty. Anxiety is an inevitable and, within limits, a healthy response to imminent surgery, for the reasons dealt with in the opening section of this paper, and only harm can come from ignoring this fact.

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Nevertheless, extreme anxiety may produce difficulties and even dangers for the nursing staff, the anaesthetist, the surgeon, and the patient himself. There is, unfortunately, little evidence to suggest that surgeons, senior nurses and hospital administrators are sufficiently aware of the anxiety-provoking potential of such "trivial" matters as sudden alterations in the time of operation, a thoughtless remark from a junior nurse or wardsman, or prolonged delay before induction in the anaesthetic room. Many patients submit to incision without ever having been made clearly aware of what surgical procedure is planned, and with only a vague and often quite incorrect notion of what results may be expected to flow from it. Even worse, they may have been submitted only a day or two earlier to the frightening experience of hearing doctors discussing, or even arguing about, their case without first moving out of earshot of the patient. The impact of ward rounds upon hospitalised patients has been recently described by Preuss et alii (1958).

## 3. The psychological significance of illness and of its possible relief by surgery

It has already been indicated that surgical procedures arouse apprehension in all patients to varying degrees because of the conscious or unconscious significance with which the operation is invested. These reactions may be

added to by various anxieties arising from the illness itself or from the associated hospitalization, especially if this has been prolonged. To those individuals whose early dependent relationship with their parents has been unsatisfying or even painful, reactivation of dependency through illness and enforced care by others may reawaken old anxieties, or at times guilt because they are ashamed of deriving satisfaction from the experience of being looked after. Such individuals, especially males, may have found the burden of supporting a family to have been an uncomfortable load, yet a role into which they have been forced by social pressures; illness may produce serious conflict, with on the one hand a sense of failure in having to abandon temporarily the task of supporting and protecting others, but at the same time a sense of relief at being able to return to their more basic dependent state with external justification. Many persons of so-called obsessional personality develop serious guilt feelings about being sick at all; however seemingly irrational these may appear, their conscientiousness and need for perfection is so great that to become sick is equated with failure and defeat.

But there is another significant group whose behaviour is in many respects the converse of the above. These are people who have suffered for many years from chronic disease and for whom new developments in surgical technique suddenly offer the prospect of partial or complete release from discomfort and invalidity. Paradoxically, such a vista and its subsequent accomplishment is not by any means always met with healthy psychological adaptation and renewed personality development, and the reasons for this reaction are usually not hard to demonstrate -many of these patients are reluctant to give up their disease, which has been invested with many neurotic gains, and which has provided them for many years with satisfying dependence and a refuge from stressful life experiences. No group has been more striking in recent years in this respect than those many patients for whom cardiac surgery, whether aimed at congenital or acquired lesions, has brought a prospect of physical well-being after years of incapacity; Kaplan (1956) and Fox et alii (1954) have both produced well-documented studies on the psychological sequelae of mitral commissurotomy. Kaplan believes that post-operative

neurotic maladjustment is so frequent that psychological evaluation is indicated in every case where this operation is performed.

Though many patients quickly capitalise upon the physical benefits of operation, some of the undesirable reactions are illustrated by the following case:

#### Case 5

A married woman of 23 had a good technical result from mitral valvotomy for a stenotic lesion, which had been first diagnosed in her early teens. Three months post-operatively however she was as incapacitated as she had ever been due to attacks of palpitations and urgent dyspnoea, with gross weakness and "nervousness", all of which settled down completely on admission to a psychiatric ward, only to recur immediately after discharge. She had always been of neurotic temperament, with some inexplicable anxieties and fears during adolescence; since her marriage at the age of 20 she had lived with her husband's relatives, and her mother-in-law had run the home and cared for the patient's only living child. Psychosexual immaturity was marked, and she had been secretly glad that her cardiac lesion gave her a sound excuse for avoiding an active sexual life. Following surgery, her alibi had gone; new responsibilities were expected of her in all areas, her husband made more normal sexual demands on her and wanted further children, and to these stresses she had no answer but to retreat unconsciously into hysterical cardiac symptoms determined by her previous organic pathology. She made good progress with supportive psychotherapy at an out-patient level, and gradually was able to leave behind her a good deal of her old dependence and inadequacy. Needless to say, there was a highly disturbed emotional background in childhood forming the basis of these developments,

There is a further group of patients to whom surgery has a special significance for deep and unique personal reasons, invariably unconscious. Every surgeon must be familiar with the occasional patient who seems actively to welcome or even to seek operations, even those of a mutilating kind; plastic surgeons in particular are dogged by this kind of individual, often seeking repeated rhinoplasty, usually determined by the well-authenticated phallic symbolism of the nose. There are also those who are unusually accident-prone, a group which is extensively described by many writers on psychosomatic medicine. Patients of this type urgently need psychiatric referral, as their motivations are inevitably deep, tortuous and complex; the following case, though a gross example of behaviour which is usually more subtle in its manifestations, typifies the problem:

Case 6

An extremely attractive young woman of 19, under treatment for a severe neurotic disturbance, contrived to swallow 7 table forks over a period of twelve months, requiring laparotomy on each occasion. Psychotherapy did not at any stage proceed far enough to unearth the obviously highly deviant motivations for such an act, but one aspect was clear enough — in the period of four to seven days following surgery, on each occasion she was cheerful and calm, and gave the appearance of radiant good health, a striking contrast to her basic state of apprehension and depression, in which compulsive eating was a feature. Whatever else might have been involved, certainly the surgical experience represented a gratifying flirtation with death and satisfied what must have been intense self-punishing self-destructive needs.

In this connection, the important findings of Beecher (1956) deserve mention. He contrasted the amount of analgesia requested by 150 male civilian surgical patients with that requested by 150 battle casualties; though the extent of the tissue damage was far greater in the latter group, the amount of analgesia requiring to be administered was much less. In drawing the obvious conclusion that pain experience has a large component of central elaboration and involves far more than mere stimulation of peripheral nerve endings, he noted that in the battle casualties the wound tended to be seen, even when severe, as leading to delivery from danger and relief from overwhelming anxiety, whereas to the civilian surgical patient tissue injury was seen as a personal disaster, an intense threat to the individual's integrity and a potential interference with his capacity to work and to support his family.

#### 4. Post-operative emotional disturbance

If we look only at the gross, unmistakable emotional disturbances which may follow surgery, often arising with dramatic urgency. then we would be justified in assuming that such sequelae of operative procedures are rare indeed. Psychosis in the post-operative period was first described by Dupuytren in 1833, but it has been since realised that "post-operative psychosis" is no special, unique entity and that any type of major mental disorder may arise at this time precipitated by the physiological and/or the psychological stress of surgery. Stengel et alii (1958), in a study of 80 cases observed during a five-year period, showed that in these patients there was no significant correlation with operative complications; furthermore, as would be borne out by any surgeon's experience, the incidence of psychosis seemed no

higher after extremely traumatic, radical procedures than after less extensive and severe operations. Though their numbers were small, they suggested — and this is of particular interest in view of the relevance of unconscious castration fantasies in surgery — that hysterectomy is more likely to be followed by psychosis than other gynaecological procedures.

But these results, as has been said, are but one of the possible sequelae of surgical operation; it must be regrettably pointed out that, in the vast majority of instances, it is only in those dramatic circumstances, where the patient is more or less "raving mad" and starts disturbing the surgeon's other patients, that his post-operative emotional status is given any real consideration. Much more frequently variations in his psychological equilibrium are of a more complex, more subtle and almost invariably completely subjective and personal kind, virtually disregarded by the surgeon and his nursing staff, but of very great importance for the patient's subsequent life adjustment.

#### Case 7

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A girl of 14 presented at the psychiatric outpatient department with a history of severe and chronic antisocial behaviour, of such magnitude as to have led to her exclusion from school. As contact was gained with her she talked freely about her envy of boys, her disgust at menstruation, and gave much evidence that she was rejecting as far as she could her feminine role. When the time came for a routine physical examination, she became anxious and hostile when requested to undress, and could only with great difficulty and many tears be per-suaded by the nurse to do so; the reason then became apparent in that her left breast was com-pletely undeveloped though the right breast was completely normal. Scar tissue existed over the left nipple region and, though the exact nature of the operation could not be clarified, it appeared that she had had some surgical procedure in early childhood for removal of a "tumour". Concerning the resultant deformity, with its limiting effects upon swimming and other activities and its possible effect upon a future husband, she was exquisitely sensitive. It is not suggested that in this case the surgeon could or should necessarily have been able to foresee the long-term emotional consequences of his therapeutic endeavours, nor is it implied that her delinquency was solely due to this cause; nevertheless, this is one of many examples in which surgical operation left a "scar" on the patient's personality and adjustment far greater than that carried on her skin,

Titchener et alii (1957) have produced some important figures bearing on this problem, based on a careful psychiatric examination on each of 200 patients before and after treatment in a surgical ward, whether submitted to operation or not. Approximately 70 per cent. of these were rated as "surgically improved" three to six months later, but one-third of these, or 23 per cent. of the total group, were emotionally more disturbed than in the pre-operation period, despite their improved status at a purely physical level. It is worth noting that patients in this group tended to be those who had passed through an unusually stormy time in the immediate post-operative period, with a good deal of anxiety and fear; similar findings in this respect have been reported by Menzer et alii (1957) in 26 patients who had a hysterectomy for benign uterine disease.

In the light of the above comments on psychological concomitants of surgery generally, it is not difficult to imagine the type of patient who falls into this important and common category of "surgically improved, but psychiatrically worse". As with the overt psychotic reactions, emotional sequelae of a less gross kind seem to bear no relation to such factors as the extent of anaesthesia, duration of surgery, amount of blood loss, and so on. Rather it is the unconscious and symbolic significance of the procedure, its repercussions upon the patient's total personality and its impact upon his life adjustment. Menzer et alii have shown in a substantial series what other psychiatrists have long since suspected, namely that it is the patient's attitude to femininity which is the prime determinant of her mental state following hysterectomy, and the psychological sequelae will be greatest in those women to whom the loss of their uterus represents an emotional catastrophe. Such reactions are of course far from confined to those who experience gynaecological surgery; other examples can be found in those who, like the case quoted above, are subjected to mutilating procedures, which are inevitably followed by some degree of depression unless this itself is dealt with by denial. "There is nothing delusional in the patient's conviction that an amputated arm or leg is an object of revulsion" (Meyer, 1958). Another example is the greater or lesser degree of emotional distress, depending on his previous level of psychological adjustment, which must inevitably result from abdominal operations which end in permanent colostomy, especially should this prove difficult to control, leading to a humiliating helplessness in dealing with excretory activities.

The frequency of such complications, even in those patients who obtain a good surgical result, is further highlighted by Lindemann (1941), who examined 40 women who had experienced uneventful somatic convalescences following major abdominal surgery; no less than 13 patients, interviewed twelve to eighteen months post-operatively, reported restlessness, insomnia and preoccupation with depressive thoughts commencing three to four weeks after the operation and lasting more than three months.

Psychomatic relationships in surgical disease

In a further report from the Cincinnati group on the 200 patients previously referred to, Zwerling et alii (1955) after full psychiatric interview, psychological tests and social histories had been obtained in each case. found that 86 per cent. of patients on a surgical ward could be shown to have a diagnosable emotional maladjustment, and in just over half these there seemed a significant relationship between the patient's current surgical status and his psychological disorder either, of course, being primary, or with both stemming concurrently from a single root. If this figure at first sight seems unusually high, the large number of possible significant connections must be remembered, such as the following:

- (a) surgical disease may be producing overwhelming anxiety, dealt with by various defence mechanisms and leading to overt psychiatric symptoms in persons of neurotic character structure;
- (b) personality disorder may have led somewhat circuitously to a surgical lesion, e.g. alcoholism leading to negligent driving and resultant trauma;
- (c) the large amount of minor or major surgery required in those patients who are accident prone (Alexander, 1952);
- (d) patients requiring surgical treatment for psychosomatic diseases, e.g. duodenal ulcer, ulcerative colitis and thyrotoxicosis, in all of which psychological stress plays a significant role.

Furthermore, a series of studies from Harold Wolff's group at Cornell University (e.g. Hinkle et alii, 1958), have shown, in several large series of cases, that there is such a pronounced difference in individual susceptibility to illness that 25 per cent. of their subjects experienced approximately 50 per cent. of all episodes of illness reported over a twenty-year period of life. Coincident emotiogenic and physical disease, as every physician knows, is extremely common.

Space permits consideration of only one aspect of this problem here, namely, the end results of surgical treatment in the so-called "psychosomatic diseases". Any general theory of such illnesses, however complex and divergent are the hypotheses regarding the underlying psychopathologic mechanisms, will emphasise that the morbid anatomical and physiological processes occuring in the duodenum, the colon, the skin, or elsewhere, are the resultants of irregular activity of the autonomic nervous system consequent upon emotional conflict or stress. In addition, then, to the psychological significance of the disease process to the patient's overall adaptation to life (as, for example, in the cases of valvular heart disease cited previously), in psychosomatic lesions an additional problem is involved; the patient's unrecognized anxiety, fear or aggression has been, often for years, worked out without his conscious knowledge on to a particular organ or group of organs. Sudden removal of this target for stress, therefore, will force the individual, still unwittingly, to deal with unresolved psychogenic conflict in a new way.

These facts have been amply demonstrated with regard to duodenal ulcer by Browning and Houseworth (1953). They examined 30 patients selected at random who had been treated by gastrectomy for duodenal ulcer, and compared their progress with 30 matched cases who had received only medical treatment, and who had been suffering an equivalent amount of symptomatic distress. Though at follow-up twelve months later only 43 per cent. of the gastrectomy group had ulcer symptoms, the incidence of other psychosomatic symptoms had increased from 13 per cent. to 37 per cent., and the incidence of neurotic symptoms proper (anxiety, depression, phobias and the like) had risen from 50 per cent. to no less than 100 per cent. In the control group, the incidence of ulcer symptoms following medical treatment was unchanged, without significant alteration in their severity, nor was there any increase in the frequency and severity of other psychosomatic or psychoneurotic symptoms (an observation, incidentally, which joins very many

others in helping to refute the popular beliefs of some surgeons and physicians that psychological symptoms in psychosomatic illness are purely the result of the lesion, rather than playing an aetiologic role.) The incidence of all types of symptoms, somatic and psychic, was 55 per cent. before gastrectomy, and 60 per cent. in the follow-up period, and the relative severity of the individual's overall distress was unchanged. A recent personally observed case may serve to illustrate the type of problem.

#### Case 8

A 30-year-old woman gave a history of dyspepsia arising originally in her adolescence in relation to parental quarrels. At the age of 20 she had a ruptured duodenal ulcer oversewn shortly after she became engaged, and this operation was repeated at the age of 24; a third rupture led to her having a partial gastrectomy when she was 27. Multiple neurotic symptoms arose virtually immediately fol-Multiple lowing this last operation; she had severe recurring depression, requiring treatment by electrotherapy on 3 occasions, developed overt anxiety and hostility to a disabling degree and became addicted to sedatives to such an extent that at one stage certification under the Inebriates Act was seriously considered. Psychotherapeutic interviews showed, as expected, that the roots of her neurosis lay deep in childhood attitudes and emotions, but both the patient and her husband stated spontaneously and convincingly that she had been completely free of overt neurotic symptoms prior to her gastrectomy.

A real need exists for further studies of this type, both in duodenal ulcer and in observing follow-up results of, for example, colectomy for ulcerative colitis or sympathectomy for hypertension. Certainly the replacement of psychosomatic by psychoneurotic symptoms has been by now noted so frequently as to be inescapable; any perceptive and sensitive dermatologist, for example, will bear eloquent testimony to this fact. It should no longer be possible to regard a gastrectomy as a "success", except in the most limited technical sense, if the patient becomes acutely neurotic or commences to take drugs six months later.

### Special problems of surgery in aged patients

The elderly person has special and important psychological problems in his own right, even if adequate physical and mental health is preserved. In our culture in particular, in contrast to certain more primitive societies, the community and family status of the old person tends to be low, and it is difficult for him in all too many instances to maintain a healthy egotism and self-esteem in the face

of repeated demonstrations of his declining usefulness to others. Personal relationships tend to be less rewarding, and social isolation may be in some instances profound; there is a partial withdrawal of interest from the outside world on to the individual's own body, i.e., an increase in narcissism, with a tendency, even in the well-adjusted, towards hypochondriacal concern and a heightened anxiety in response to even minor defects in the body image, including any awareness of failing mental efficiency. All these shifts in the psychological balance of forces will be aggravated in those individuals whose previous personal relationships have been unsatisfactory because of neurotic self-centredness, or for whom their life situation in their declining years fails to be supportive and protective.

The sequelae of surgical procedures in the aged patient, in contrast to the younger group, are to a significant extent determined by factors related to organic cerebral damage. Titchener et alii (1958) found that, in 45 patients aged 65 years or over submitted to surgery, no less than 11 showed post-operative mental changes of an organic type, in 7 of which the permanent and progressive deterioration had been preceded by an acute delirium. Patients aged 70 or over appear more likely to develop this most serious type of complication. In a larger series (Bedford, 1955), relatives described a change in mental function in one-third of the group; "substantial evidence of brain damage" was found in 120 out of 1,193 patients, with 30 of these showing "severe cerebral damage". It is usually impossible in such cases to locate the exact causative factor, and the mental deterioration can only rarely be positively attributable to the duration and degree of hypotension or the extent of blood loss. In most instances one must assume that the individual comes to surgery with minimal brain cellular damage adequately compensated, and that comparatively minor degrees of further damage due to hypoxia, together with the psychic stress of the surgical procedure, tips the scales to produce a decompensated state of mental confusion and disorganization.

But even more common than these organically based reactions are the psychiatric sequelae which appear in the presence of clear consciousness and which are determined, but with added frequency, by essentially similar factors to those described earlier in the

paper. In addition, the factors surrounding hospitalisation itself and the inevitable separation from familiar faces and the patient's customary environment and mode of life, seem on the basis of ordinary observation to play a larger part in the emotional reactions of the senescent individual. It is exceedingly common to see a flat, pessimistic, bitter depression in such patients in a surgical ward; florid manifestations of psychosis, as in younger people, are extremely unusual but the "will to live" is often weak or absent, especially following the death of another patient in the ward. Titchener et alii (1958) suggest that the cheerlessness and barrenness of most hospital wards, the occasional impatience of hospital personnel with the old person's slow reactions and fixed habits, and long periods of immobilization, all must be factors which help to initiate or accentuate this process. In the assessment of the significance and management of these common and important reactions, it would be hard to lay too much stress on the relevance of the patient's social, material and personal environment and impossible to overestimate the value of a skilled almoner in their handling. It is also noteworthy that in this age group surgical procedures are especially liable to lead to depression and hopelessness should they involve loss of a body part which will curtail activity.

#### 7. Surgery in childhood

One still hears it said at times that the psychological and emotional immaturity of the young child tends to protect him from any undue psychic reaction to the stress of surgical operation. Nothing could be more illinformed or more dangerous, for in fact the very opposite is true; it is the child's primitive type of thinking, his ready recourse to fantasy, and the generally turbulent and changeable state of his emotions which makes him peculiarly susceptible to severe personality deformity as a consequence of operative procedures which he but dimly understands, if at all, and for which he has all too often been inadequately prepared. Psychoanalytic observations, such as reported by Anna Freud (1952), make it clear that, contrary to much popular opinion, the child's capacity to cope with unavoidable external traumata is much increased by adequate psychological preparation and foreknowledge up to the limits of his capacity to understand, a task which many parents (and surgeons) still frequently avoid

by reason of their own anxiety. Findings of this nature have greatly influenced the techniques of such surgeons as Lester Coleman, a New York oto-rhino-laryngologist, who "insists that the child comes to see him two or three times before the operation, in his office. He plays with them, tells them all about the hospital and what is going to happen. They play with the anaesthetic apparatus. In the hospital . . . he always fetches the child himself from the ward, because it gives him time to talk while they walk upstairs. . . . Always during this stage the child brings up her greatest anxiety, usually anxiety about un-consciousness. 'And shall I wake up?' He sits down on the stairs and talks about this before they go on to the operating theatre; he is prepared to talk about it for five minutes" (Hargreaves, quoted in Tanner and Inhelder, 1956).

The additional unique factor in childhood illness is the mother-child separation which is brought about by hospitalisation, the importance of which has been stressed by Bowlby et alii (1956). Further studies require to be done to confirm and elaborate the long-term effects of separation on personality development, but in the meantime more than enough evidence has been produced to make it quite clear that even a temporary interruption of continuity in the closeness of mother and child, especially in the first two to three years, should be avoided whenever possible. Viewing of the film, "A Two-Year-Old Goes to Hospital" (Robertson, 1953), should be compulsory for all paediatric surgeons; perhaps a long overdue result of this might be the performance of more surgery on children on an "admission and discharge" basis, or even in an outpatient theatre, avoiding some of the many traditional and inappropriate rituals associated with the admission of children to hospital.

#### SUMMARY AND CONCLUSIONS

- 1. Consideration of patients as whole persons functioning in a certain social environment, rather than as aggregates of diseased or healthy organs, requires an understanding of certain basic tenets of modern dynamic psychology. The concepts of unconscious motivation, the body image, narcissism and castration anxiety are vital in this connection.
- 2. All surgical procedures must inevitably arouse some degree of anxiety; the manner in which this is dealt with, both by patient and

surgeon, will frequently have profound effects on the individual's total adjustment. Failure to recognise anxiety may vitiate an otherwise successful technical result.

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- 3. Many surgical patients can be helped by psychiatric assessment and by brief psychotherapy, either in the pre-operative or post-operative periods or both.
- 4. Surgery in the aged poses special psychological problems, which may be substantially alleviated by close study of the patient's personal, social and environmental situation. At this period of life post-operative toxic-confusional states lead so frequently to irreversible mental deterioration that every effort must be made by surgeons and anaesthetists to avoid their occurrence.
- 5. Surgery in childhood represents an important and potentially very harmful emotional stress; elective operations should be postponed as long as possible, and admission to hospital recommended only when strictly unavoidable.
- 6. There are enormous potentialities for future research in the areas of interrelation-ship between psychiatric and surgical practice. For this reason, in addition to the implications for therapy of such a move, there is a real and urgent need for the provision of more adequate psychiatric consultative services within general hospitals, including the appointment of a liaison psychiatrist to any surgical service of reasonable magnitude.

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#### VESICAL DIVERTICULA IN CHILDREN\*

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FOR many years there has been considerable diversity of opinion concerning the aetiology of diverticula of the bladder. Two types of vesical diverticulum have been generally recognized; the congenital, which exists from birth as an anomaly of development and the acquired which develops after birth, presumably the result of urinary back pressure.

The majority of authors who have studied this subject consider the primary cause to be urethral obstruction, with the diverticula appearing secondarily. So widespread is this view that the presence of a diverticulum of the bladder is considered to be almost diagnostic of urethral obstruction.

#### MATERIAL AND METHODS

In the years 1951 to 1958, 23 patients exhibiting diverticula of the bladder have been studied in the Urological Clinic of the Royal Children's Hospital in Melbourne. In 13 patients diverticula were associated with bladder neck or urethral obstruction; in 10 patients, however, no obstructive factor could be detected. The 10 children comprising this group are considered in detail in this survey. These patients were all subjected to full routine urinary investigation, supplemented by micturition cysto-urethrography. In four patients, electro-manometric voiding pressure studies of bladder and urethra gave additional information.

Table 1
CLINICAL FEATURES OF TEN PATIENTS EXHIBITING VESICAL DIVERTICULA

Patient	Sex	Age when first seen	Duration of symptoms when first seen	Urinary Tract Infection	Dysuria	Enuresis	Positive D.M. test
K.W.	M.	8 yrs. 10 mths.	3 years	+	-	-	+
B.B.	M.	8 yrs. 2 mths.	3 days	+	+	-	Not tested
A.C.	M.	13 yrs. 7 mths.	10 years	+	+	+	Not tested
R.S.	F.	3 years	1 year	+	+	-	+
P.W.	M.	8 years	3 years	+	+	+	+
A.D.	F.	11 years	1 yr. 6 mths.	+	+	-	Not tested
P.S.	F.	7 yrs. 7 mths.	2 months	+	+	-	Not tested
S.H.	F.	8 yrs, 5 mths.	1 year	+	+	+	Not tested
H.K.	M.	7 years	5 years	+	+	+	+
M.Y.	F.	7 years	2 years	+	_	+	Not tested

In our study of vesical diverticula in children, it was found that obstruction of the urethra was not the only cause. In this paper we record our observations and conclusions regarding (1) aetiology of the diverticula, and (2) the relationship of the diverticula to vesico-ureteral reflux.

#### CLINICAL FEATURES

The clinical features of the 10 patients are summarized in Table 1. Five were male and five female. The duration of symptoms varied from three days to ten years.

Urinary tract infection was the chief presenting feature in all patients. One patient experienced only one episode; the remainder exhibited three or more attacks.

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Pain with micturition was present in 8 patients. In 6 of these, dysuria occurred in relation to an attack of cystitis and urethritis; in two, pain with micturition occurred in the

FIG. I. Vesico-ureteral reflux is proportional not to size of diverticulum but to intimacy of the orifices of diverticulum and ureter.

- (a) K.W.-Orifices adjacent: no reflux.
- (b) A.C.—Orifices contiguous: limited reflux (arrows).
- (c) R.S.—Ureteric orifices engulfed by large right and small left diverticula: free reflux.

(Micturition cysto-urethrography series— (a) and (b) voiding and (c) at end of voiding showing bladder empty but both diverticula distended with iodide.)

absence of infection. Here it was associated with distension or overdistension of the bladder, intense pain accompanying the onset of micturition and discomfort remaining for a minute or two afterwards.

Enuresis was an incidental feature of five patients.

#### Double micturition test (D.M. Test)

By the act of micturition in normal children, the bladder is completely emptied and remains so for the next few minutes. If the child can pass a second quantity of urine two minutes later, the presence of residual urine in the urinary tract is suspected. This residual content may reflux into the ureters, or into a vesical diverticulum or it may be

retained in the bladder if the urethra is obstructed. Its location can be determined by micturition cysto-urethrography.

In four patients only this test was applied and in all four the finding was positive, with respect to the presence of residual urine.

#### RADIOGRAPHIC FEATURES

In this series all the children were examined by micturition cysto-urethrography. The test did not supplant the standard radiographic techniques but was used in addition to them. Cysto-urethrography is important not only for diagnosis, but also for studies of the physiological activity of the diverticula and of other parts of the urinary tract.

In this series, special observations were made on the diverticula, the ureters and the urethral outflow.

#### (a) Diverticulography

In normal children, the bladder outline during voiding becomes finely irregular. Sometimes these irregularities over the posterior wall near the bladder base assume the dimensions of flat-roofed sacculations which may be single or multiple. The bladder outline usually retains a rounded shape but the thin lateral walls may bulge during micturition thereby creating a trilobar outline when the vesical outline is viewed in the antero-posterior projection. These are variants of normal, the bladder empties completely and they create no symptoms.

The shape and size of diverticula were different when viewed during the resting and emptying phases of bladder activity. Small diverticula or even inconspicuous small irregularities in bladder outline ballooned into large and rounded protrusions during micturition. The diverticula remained filled for half a minute after micturition; the fluid then spilled back into the bladder.

Diverticula in these children were found to assume two shapes: Firstly the globular protrusion, unilateral or bilateral, commonly near the trigone, often visible radiographically only when distended during micturition or when filled at the end of micturition (Fig. I). In two children, similar local isolated protrusions occurred elsewhere on the posterior wall. Secondly, and less commonly,

the lateral bulges of the bladder wall were exaggerated. In addition, they were studded with sacculations and the effect produced was of large irregular lateral herniations of the bladder wall. During micturition, these lateral weaknesses of the bladder and the superimposed sacculations were even more prominent (Fig. II).



FIG. II. P.S.—Diffuse wide neck diverticulum.

Micturition cysto-urethrogram showing the bulge
in the lateral wall of the bladder.

#### (b) Ureterography

In five patients, reflux was noted. In one patient, this was bilateral and in the remainder it was unilateral.

The reflux was free in four ureters which were regarded as larger than normal in calibre. Limited reflux—a fine iodide streak only was visible—occurred in two other ureters, both of which were normal in calibre (Fig. I).

In one other patient, H.K., free ureteric reflux occurred up the left ureter, though a large herniation type diverticulum lay in the right wall of the bladder.

#### (c) Urethrography

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A careful search was made of all voiding cysto-urethrograms to detect possible sources of urethral obstruction. Neither the bladder neck nor the urethra could be incriminated.

The bladder neck opened widely during voiding; the calibre of the urethra in both males and females was within normal limits (Fig. I).

In our view, these radiographic appearances of the bladder neck and urethra in the state of physiological expansion during micturition, were sufficient to eliminate obstruction as a primary cause of the vesical diverticula. Further confirmation was obtained in four children by electromanometric methods which were combined with these radiographic techniques.

In girls, there is normally little or no difference in urethral and bladder pressures during voiding. In boys a fall in pressure is from 10.0 to 15.0 mm. of mercury between bladder and bulb is accepted by us as being within normal limits.

In the two girls, simultaneous pressure recordings in the bladder and urethra, both at the onset of voiding and at the time of occurrence of the maximum pressure, were the same. This indicated that there was no obstruction at the bladder neck. In one boy, the vesical and urethral voiding pressures were normal. In the other boy, it was impossible to obtain simultaneous pressure recordings in the bladder and bulb. He could not tolerate the second catheter in the urethra. The height of pressure in his bladder at the

Table 2
BLADDER AND URETHRAL PRESSURES IN FOUR PATIENTS EXHIBITING VESICAL DIVERTICULA

Initials	Sex	Age in years	Resting pressure in bladder	Bladder pressure at onset of voiding	Urethral pressure at onset of voiding	Maximum pressure in bladder	Maximum pressure in urethra	Duration of voiding
P.W.	M.	131	10 mms.	24 mms.		43 mms.		30 secs.
A.C.	M.	12	8 mms.			48 mms.	36 mms.	25 secs.
S.H.	F.	91	9 mms.	26 mms.	19 mms.	33 mms.	33 mms.	19 secs.
P.S.	F.	11	6 mms.	14 mms.	9 mms.	25 mms.	25 mms.	9 secs.

Simultaneous recordings of bladder and urethral pressures during voiding. Catheter tip in bulbous urethra in males and at a point 1.0 cm. inside external urethral orifice in females. Pressures recorded in millimetres of mercury.

#### ELECTRO-MANOMETRIC STUDIES

Electro-manometric pressure readings of bladder and urethra were undertaken in two girls and two boys. Of these, one was studied before operation, and three have been studied after excision of the diverticula (Table 2).

The pressures were recorded in the bladder and urethra simultaneously using a Southern Twin Channel capacitance electro-manometer. In both boys and girls, one fine catheter (size No. 5 cardiac) was introduced per urethram into the bladder. A second catheter of similar gauge lay approximately 1.0 cm. inside the external urethral orifice in the girls, and in the bulbous urethra in the boys.

onset of voiding (24.0 mm, mercury) and the maximum pressure during voiding (43.0 mm, mercury) indicated that normal intravesical pressures were required to initiate and maintain voiding (Table 2).

From these recordings, it is presumed that urethral obstruction is not the cause of the formation of the diverticula.

These studies have shown that two types of diverticula, of non-obstructive aetiology, can be identified radiographically, viz., the local globular pocket and the diffuse lateral bulge of the bladder. Further information concerning the nature, shape and location of the two types was obtained by direct vision.

#### CYSTOSCOPIC AND OPERATIVE FINDINGS

These observations were made at cystoscopy and at open operation on the bladder. Two types of diverticulum could be distinguished according to the dimensions of the neck:

- (a) the narrow neck globular protrusion,
- (b) the wide neck lateral herniation.

The locations of the diverticular orifices in this series are shown in Fig. III. They are grouped mainly around the ureteric orifices but they also appear in other areas of the bladder. Fig. IV shows the relation of the diverticular orifice to the ureteric orifice in 8 of our patients. In the other two patients, the diverticular orifices were placed too far away from the trigone to be conveniently included in this diagram. In 7 patients, the diverticula were single, in two they were bilateral, close to each ureteric orifice, and in one they were multiple. The study of diverticula has made us think in terms of subdividing them into groups according to their appearance by cystography and at cystoscopy.

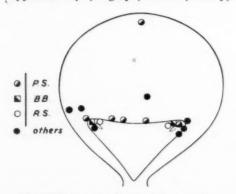


FIG. III. Distribution of diverticula in bladder.

Note the preponderance around the trigone.

(Black dot represents solitary diverticulum.)

#### (a) Narrow neck diverticula

These diverticula were globular in shape and joined the bladder through a narrow neck. This was the common variety and occurred mainly in relation to the lateral cornu of the trigone sometimes separate from, sometimes impinging on and sometimes engulfing the ureteric orifice. They were unilateral, or bilateral, and in one patient they were associated with pea-sized saccules in

other areas of the bladder. Less commonly the diverticulum may lie elsewhere in the wall of the bladder.

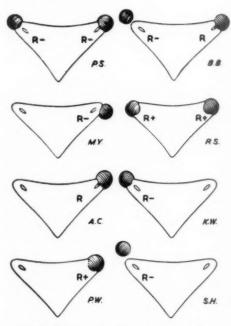


FIG. IV. Trigones of 8 patients to show relationship of location of diverticulum to ureteric orifice and to vestico-ureteral reflux in each. (R- denotes no reflux: R denotes slight reflux: R+ denotes free reflux:

#### (b) Wide neck diverticula

In this group, the rare variety, the appearance suggested a wide herniation of the lateral wall of the bladder (Figs. II and VIa). The protrusion was wider at its neck than at the dome. The diameter of the neck was 3 to 4 cm. The actual diverticulum was difficult to discern cystoscopically but at operation the thin bladder wall and its sacculations were demonstrable as pockets into which the finger tips could be inserted and between which firm cords of trabeculae could be identified. Three children, S.H., P.S. and H.K., exhibited this variety (Tables I and 3).

#### CORRELATION OF RADIOGRAPHIC AND ANATOMICAL FEATURES

Our cystoscopic and operative findings when correlated with the results of radiographic studies, showed clearly that the narrow and wide neck types corresponded with the globular and diffuse protrusions respectively. Furthermore, they revealed an interesting relationship between the location of the diverticular orifice and competence of the uretero-vesical valve.

When the locations of these orifices were plotted they fell into three groups (Fig. V):

- Group 1: The ureteric and diverticular orifices though closely related were separate and in these there was no reflux.
- Group 2: The ureteric orifice was contiguous with the orifice of the diverticulum; this orifice could be visualized and catheterized. In these two there was only slight reflux and no ureteric enlargement.
- Group 3: The orifice of the ureter in three instances was engulfed by the diverticulum and could not be seen nor catheterized. There was free reflux and the ureter was enlarged in calibre.

In one patient, a right-sided diverticulum completely separate from the right ureteric orifice, showed reflux up the opposite left ureter.

#### STRUCTURE OF DIVERTICULA

Eleven specimens of diverticula, excised at operation, were available for histological examination. Eight of these were the narrow neck globular type and three were diffuse diverticula. Both haematoxylin and eosin and Masson stains were used to identify muscle fibres and fibrous tissue.

Of the eight narrow neck specimens, fibrous tissue but no muscle, could be found in one only. In seven specimens, one or more muscle bundles traversed the walls of the diverticulum and fibrous tissue filled the wide gaps between these fine cords of muscle.

In the three specimens of the wide neck type of diverticulum, numerous muscular trabeculae were identified in the thin wall of the diverticulum. These individual cords of muscle were not as thick as the wall of a normal bladder. Between these cords, there were deficiencies which were filled in by fibrous tissue. The individual muscle fibres in the diverticulum were large suggesting some degree of hypertrophy.

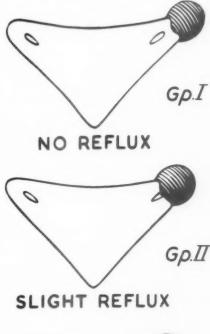




FIG. V. Correlation of intimacy of orifices of diverticulum and ureter to vesico-ureteric reflux.

Macroscopic inspection of the bladder wall adjoining the narrow neck or the diffuse diverticula, revealed a sudden return to normal thickness. From within the bladder, the deficiencies presented smooth selvedged edges.

#### AETIOLOGY OF DIVERTICULA

For descriptive purposes vesical diverticula may be divided into two groups:

- (a) The group associated with primary obstructive pathology in the bladder neck or urethra.
- (b) An idiopathic group in which the urethral outflow is unobstructed.

Most authors (Kretschmer, 1934; Miller, 1958; Hinman, 1919) have concluded that bladder neck or urethral obstructions are primary factors in nearly all cases of vesical diverticula. However these conclusions have been based on the observations of series consisting of large numbers of diverticula in adults and very few in children.

Hinman (1923), Badenoch (1949), Campbell (1951) and Williams (1958) report 9 cases of diverticula which they consider were unrelated to urethral obstruction.

Many theories have been put forward to explain the aetiology of idiopathic diverticula of the bladder. Campbell (1951) summarizes them as follows:

- "(a) Retention in fetus due to temporary occlusion of the urethral mucosa (Englisch).
- (b) A superabundance of embryonic tissue in the bladder wall.
- (c) The formation of excess of epithelial tissue at the fusing edges of the wolffian and allantoic elements of the bladder and a temporary failure of epithelialization between the two.
- (d) Supernumerary buds.
- (e) Patent urachus, the probable cause of all cases found at the dome."

The theory has also been put forward by the German urologists, Auschuetz, Lurz and Blum (quoted by Kretschmer, 1934), that diverticula are not present at birth but are foreshadowed by a muscular weakness in the bladder wall. This theory would seem to postulate a congenital anlarge of the diverticulum. With the development of urinary obstruction or even the normal force of urination, the diverticulum may appear either antenatally or after birth. Areas of muscular weakness in the wall of the bladder have been histologically demonstrated by Marsella (quoted by Kretschmer, 1934) and more recently by Uhlenhuth (1953), while Rose (quoted by Kretschmer, 1934) furnished a

histological study demonstrating the presence of unprotected loose fibrous connective tissues in the wall of the bladder.

In all our cases, except one, muscle fibres were demonstrated in the wall of the diverticulum, but the amount of muscle, though variable, was much less than is found in the normal bladder wall. These areas, defective in muscle, were found principally in the region of the ureteric orifices, though some lay on the lateral and posterior walls more distant from the orifices. We believe that a defect in the muscle coat is the primary developmental anomaly and that herniation of the bladder mucosa takes place between the muscle bundles as a result of the normal bladder pressure associated with micturition both during intra-uterine life and after birth. These diverticula may therefore be present at birth or develop gradually during infancy and childhood.

Consequent on our microscopic study, it appears that both the narrow neck and the wide neck diverticula have a similar pathology, namely congenital hypomuscularization, a failure of normal muscle development, of an area of the bladder wall. This lack of muscle may be of varying degree. Where it is almost complete and small in area, a narrow neck globular type of diverticulum develops with mucosa and little or no muscle tissue in its wall. This type of diverticulum is found most commonly in the region of the ureteric orifices. Where the failure of muscularization is more extensive, the wide neck type of diverticulum develops. This type was seen in our patients on the lateral walls (Figs. II and VIa) occupying that area of the bladder wall which normally lacks the longitudinal coats being composed of circularly disposed muscle only (Uhlenhuth, 1953). Hypomuscularization of the circular coat accounts for this wide neck diffuse type.

Clinical study of our 10 cases, convinces us that these diverticula are entirely unrelated to urethral obstruction.

Furthermore, we believe that diverticula form as a consequence of obstruction only if there are weak areas in the walls of the bladder.

#### TREATMENT

Of the 10 patients in this series, 9 have been subjected to surgery. The tenth child was managed conservatively as the diverticulum was small and there was no associated reflux. Recent radiographic studies have, however, shown an increase in the size of this diverticulum and there have been further episodes of infection; this child may yet, therefore, require surgery.

The indications for surgery were:

- 1. Recurrent urinary tract infection, inadequately controlled by chemotherapy.
- 2. Painful micturition.

Persistent infection in the urinary tract was due either to associated vesico-ureteral reflux or to the retention of residual urine in the bladder consequent on inefficient expulsion.

For the narrow neck type of diverticulum orthodox methods of surgical repair have been used, while for the wide neck type a different method was employed.

Orthodox repair

The orthodox method consisted of dissecting the diverticulum in the extravesical tissues, from the fundus to its neck. In the para-ureteral diverticula, care was exercised in the separation of the diverticulum from the ureter. The diverticulum was inverted into the lumen of the bladder, drawing the ureteral orifice with it. The neck of the diverticulum was then divided, leaving a protective margin of mucosa adjacent to the mucosal layer of the bladder were repaired in separate layers with catgut stitches (Young and Davis, 1926).

This surgical technique has been applied to 9 diverticula in 7 patients.

The unorthodox repair

Two diverticula of the wide neck type were treated surgically by a type of repair somewhat similar to the Mayo repair of umbilical hernia.

Instead of excision, the thinned out muscle coat was incised along its greatest diameter. With several fingers of one hand within the bladder, the diverticulum was made to bulge outwardly. An incision was then made through the wall of the diverticulum down to but not through the mucosa, which was then separated from the diverticulum. The walls of the diverticulum were overlapped as far as the edges of the muscular deficiency. These overlapping muscular flaps were then anchored in position with interrupted mattress sutures of chromic catgut. The excess mucosa

was then trimmed away from within the bladder. The mucosal margins were coapted with a continuous plain catgut suture (Fig. VI).

A hydronephrotic kidney, its ureter and part of the associated diverticulum were removed in one patient, P.W. Here the chief indication for radical surgery was the derelict kidney.

#### RESULTS OF TREATMENT

Operations have been performed on 9 patients. Eleven diverticula have been repaired. One patient who still has symptoms has declined operation.

These patients have been followed for periods ranging from six months to two years since operation.

All are in good health. They are symptom free and in all but one the urine is sterile.

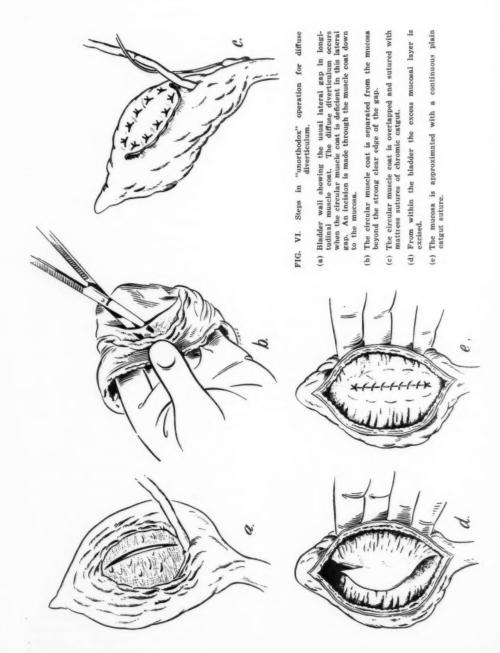
Because of vesico-ureteral reflux, the triple micturition regime is required in six patients. Two of these patients, including a child on whom the "unorthodox" operations were performed, were troubled initially with urinary infections until the triple micturition regime (Stephens, 1957) was invoked. Now, only this one patient, after the unorthodox repair, is still troubled by cystitis.

The effect of excision of the diverticulum and repair of the bladder wall on vesicoureteral reflux was variable (Table 3). The results concerning individual ureters are as follows:

- (a) Reflux cured by operation \_\_\_\_ 2
- (b) Reflux persisting after operation 3
- (c) Reflux occurring after but not before "unorthodox" operation ....
- (d) Reflux unrelated to diverticulum 1

It is our belief that vesico-ureteral reflux is not harmful to kidney function in the absence of infection or urethral obstruction (Stephens, 1956). We have indicated that neither factor operates in these patients. Triple micturition, a simple manoeuvre, practised once daily, is a measure which must be adopted to prevent infection.

This expedient, in the presence of a vesical diverticulum, is inefficient. It will not totally remove the residual urine and so infection persists. Once the diverticulum is removed, however, triple micturition becomes efficient, and infection can be prevented.



EFFECT OF DIVERTICULECTOMY ON VESICO-URETERAL REFLUX AND CLINICAL STATE TABLE 3

Present status	Well. No further urinary tract infections	Very well. No further infection. No pain	Well, Urine free from infection, Using triple micturition. No pain	Very well. Doing triple micturition. No pain	Diverticulum only partially removed	Very well. No pain	Recurrence of infection. Triple micturition started. Urine now sterile, No pain	Very well. Free of infection. No pain	Recurrence of infection. Triple micturition therapy. No pain	Recurrent infections
Reflux after operation	Slight	R. Nil	Free	R. Nil L. Free	Reflux into diverticulum	Nil	R. Free L. Free	NII	Reflux—left ureter	
Reflux before operation	Nil	R. Nil L. Slight	Slight	R. Free L. Free	L. Free	Nil	NII	N	Reflux—left ureter	Nil
Operation	Orthodox excision	Orthodox excision of diverti- cula and repair	Orthodox excision and repair	Orthodox excision and repair	Nephro-ureterectomy and partial excision of diverti- culum	Orthodox excision and repair	Overlapping repair	Orthodox excision and repair	Excision	No operation
Type of diverticulum	Narrow neck (Fig. IV)	Narrow neck (Fig. IV)	Narrow neck (Fig. IV)	Narrow neck (bilateral) (Fig. IV)	Narrow neck (Fig. IV)	Narrow neck (posterior wall)	Wide neck (bilateral) (Fig. IV)	Wide neck (Fig. IV)	Wide neck	Narrow neck (Fig. IV)
Length of follow-up	3 yrs.	1 yr. 2 mths.	1 yr. 2 mths.	1 yr. 4 mths.	2 years	1 yr. 10 mths.		13 mths.	6 mths.	9 years
Age at operation	10 yrs. 8 mths.	8 yrs. 6 mths.	13 yrs. 9 mths.	9 yrs. 9 mths.	13 years	11 yrs. 6 mths.	12 years	8 yrs. 6 mths.	8 yrs. 2 mths.	16 years
Patient	K.W.	B.B.	A.C.	R.S.	P.W.	A.D.	p.s.	S.H.	H.K.	M.Y.

The relationship of the diverticulum to vesico-ureteral reflux and the effects of diverticulectomy upon the reflux are sufficiently interesting to merit further discussion.

#### VESICO-URETERAL REFLUX

Vesico-ureteral reflux is an abnormal phenomenon which is not uncommonly observed in children. It is the result of a debilitated valve in the distal end of the ureter. The reflux causes stasis which is the precursor of infection. It may occur in one or both ureters, or in multiple ureters. It is commonly found in paraplegic patients. In this paper we have described its existence in association with diverticula of the bladder.

The association of vesico-ureteral reflux with para-ureteral diverticula is well recognized. Interference with the mechanics of the uretero-vesical junction by the diverticulum is the popular explanation of reflux (Hutch, 1959). Other possibilities should however be explored. Could the explanation be an intrinsic malformation of the ureter which shares a common aetiology with that of the diverticulum?

We have already indicated that we believe that the cause of the diverticula in the non-obstructive group is a malformation of the bladder wall — a localized total or partial lack of muscularization of the bladder wall. Perhaps this same lack of muscularization may affect those ureters whose orifices are encroached upon by the diverticula.

None of the non-obstructive diverticula with the adjoining bladder and ureter was available for examination. But in order to investigate this hypothesis, microscopy studies were made on 2 obstructive diverticula and their "swallowed" ureters. Both these diverticula and the megaureters co-existed with severe obstructions of the urethra caused by urethral valves. Vesico-ureteral reflux occurred into the diverticula and the ureters.

Paraffin blocks were made of the diverticula and the adjoining bladder wall and ureter of these two specimens. Serial sections were cut at  $15\mu$  thickness and stained with Masson stains. Similar blocks and sections were obtained of 10 normal ureters and of 2 hypertrophic megaureters and their adjoining bladder musculature.

The diverticula were compared with the biopsy material of the non-obstructive types; their ureters were compared with the 10 normal ureters and with the 2 hypertrophic ureters. These findings were most interesting.

The walls of the diverticula showed progressive thinning of the muscle coat from the neck to the dome where the walls were in part deficient in muscle.

The ureteric orifice lay inside the neck of the diverticulum in each specimen. An attenuated band of fine muscle fibres of the trigone could be traced through the neck to the ureteric orifice. The ureter almost immediately took an oblique course through the medial and posterior walls of the diverticulum. The muscle coat of the ureter was so fine in one specimen that the ureter resembled in its intramural course an epithelial lined tube. In the other specimen, an abnormally thin muscle coat was identified. The lumina of both ureters were larger than normal.

In both these specimens the primary obstructive pathology lay in the urethra: the vesical diverticula and the enlargement of the ureters and the reflux were regarded as secondary to it. If this was the true explanation, one would expect to find hypertrophy of the ureteric musculature. But the muscle of the uretero-vesical segments of these ureters was considerably less than the thickness of that of the same segments of 10 normal ureters. An even greater contrast was apparent when the muscle thickness was compared with that of the terminal segments of the two ureters which were hypertrophied secondary to urethral obstruction, but which did not exhibit diverticulum formation or reflux.

The muscle of these two ureters at the level of the pelvic brim was thicker but here again it was not as thick as the muscle coats of the uncomplicated obstructive megaureters.

We believe then that the diverticulum, the ureter within it and the ureter cranial to it, are all hypomuscular and that the hypomuscularity of the uretero-vesical valve accounts for the reflux.

These arguments could reasonably apply to the non-obstructive variety. We have shown that the diverticula are lacking in muscle and we now presume that the engulfed ureterovesical segment of the associated ureter shares in this pathology. In these engulfed ureters, this pathology may account for the ureterovesical reflux.

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Variations in the degree of muscularization of the terminal uretero-vesical valve segment may account for the variable effects of diverticulectomy on reflux. It would also explain the limited "streak" of reflux which occurs when the ureteric orifice is contiguous with but not inside the lip of the diverticulum, and the very free reflux which is invariable when the ureteric orifice lies within the diverticulum (Figs. I and V).

When the ureteric orifice was adjacent to but not contiguous with the orifice of the diverticulum, reflux did not occur. It was found at operation that the body of the diverticulum and the ureter were contiguous and traversed the same intramural tunnel. Here again it is presumed that the ureteric wall contains its muscular uretero-vesical sphincter mechanism and that the diverticulum though close does not impede its function.

Finally, in patient H.K., the diverticulum occurred on the contralateral side to that of the vesico-ureteral reflux. On this theory, reflux could be explained by local hypomuscularization of the uretero-vesical segment of the ureter.

#### SUMMARY

- Twenty-three patients with diverticula of the bladder were treated during an eightyear period at the Royal Children's Hospital, Melbourne. In 13 patients, the diverticula co-existed with proven urethral obstruction. In 10 patients the diverticula were not associated with an obstructive factor. These 10 patients and their diverticula are described in this paper.
- 2. The aetiological factor responsible for the formation of the diverticula unassociated with obstruction of the urethra, is a localized developmental muscular defect of the bladder wall. The diverticulum itself is a protrusion caused by the normal ranges of intra-vesical pressure exerted on the bladder wall in the course of normal function.

Two types of diverticula were identified—the localized globular pocket with narrow neck, and the less common diffuse wide necked bulge of the lateral wall of the bladder.

- The relationship between diverticula and the occurrence of vesico-ureteral reflux is discussed. Reflux is related not to the proximity of the body of the diverticulum to the ureter wall nor to the size of the diverticulum but to the intimacy of their orifices.
- 4. The view is put forward that the anomaly of muscular development which results in diverticulum formation also involves the terminal ureter and is responsible for impairment of the intrinsic uretero-vesical valve mechanism.
- Orthodox techniques were used to repair the globular narrow neck type of diverticulum, but an overlapping repair was invoked for the wide neck variety.

#### ACKNOWLEDGEMENTS

We wish to thank our colleagues of the Senior Medical Staff of the Royal Children's Hospital, Melbourne, for referring these patients to us for study.

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#### CONGENITAL LOBAR EMPHYSEMA\*

By N. A. MYERS

Melbourne

EMPHYSEMA of part or whole of the lungs in children has a diverse aetiology. The cause may be infective, mechanical or secondary to spasm and in some cases there appears to be little doubt that the condition is congenital. Under these circumstances, the disease process is usually confined to one or other lobe. Various names have been suggested to describe regional emphysema of this type; of these, the term congenital lobar emphysema is now used widely and is probably the most satisfactory.

Congenital lobar emphysema is usually a serious condition and may prove to be fatal; this is particularly true in infancy. Lobectomy is the treatment of choice and in the majority of cases proves to be curative. The purpose of this paper is to draw attention to this condition, to discuss certain of its aspects and to report a further case.

#### CASE REPORT

F.F., a female baby aged 7 weeks, was admitted to the Royal Children's Hospital, Melbourne, in April, 1958. The main symptoms were feeding difficulties, constipation and failure to thrive. On the day of admission, the baby had developed a respiratory tract infection. Examination revealed a dyspnoeic baby with evidence of bronchiolitis and an X-ray of the chest showed emphysema of the right upper lobe (Fig. I). Following control of the infection, right upper lobectomy was performed. At operation the lobe was seen to be voluminous and pale, and did not collapse when the chest was opened. The right middle and lower lobes were almost completely collapsed. An uneventful postoperative course followed and a subsequent X-ray (Fig. II) revealed a satisfactory state of affairs. The pathologist reported on the resected specimen as follows:—

"The bulk of the lobe is emphysematous. There is a wedge of collapsed tissue along one border which has the appearance of unexpanded lung (true atelectasis). Section of several bronchi reveals cartilage in their walls in normal amounts although the thickness of the muscle coat is reduced. Two of the larger bronchi contain mucus which occupies a considerable portion of their lumina. Mucosa in most bronchi is abundant and folded."

This case illustrates the main features of congenital lobar emphysema in infancy. Tachypnoea amounting to dyspnoea is the cardinal symptom; mediastinal displacement accompanied by an area of diminished air entry and a hyper-resonant percussion note are the typical physical signs and radiologically there is an area of increased translucency, which still shows broncho-vascular markings.



FIG. I. X-ray of chest of baby age seven weeks, showing emphysema of the right upper lobe.

In infancy there is little doubt that once the condition of congenital lobar emphysema is diagnosed, lobectomy should be performed relatively soon.

DISCUSSION

<sup>\*</sup>Received for publication, 28th August, 1959.

There has been considerable discussion regarding the aetiology and the frequency of association of congenital lobar emphysema with congenital heart disease has been noted (Cottom and Myers, 1957). In many cases deficiency of bronchial cartilage has been observed; in others a valvular effect secondary

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FIG. II. X-ray of chest following upper lobectomy. (Same case as Fig. I.)

to redundant mucosa has been described. Other aetiological factors which have been suggested include the effect of respiratory resuscitation in the new born period and the role of infection. However, in most of the cases reported so far, the evidence suggests the cause is congenital rather than acquired. For this reason the use of the term congenital lobar emphysema would seem to be justified (Potts, 1958). Other terms which have been suggested from time to time include localized hypertrophic emphysema (Kirklin et al, 1956), tension emphysema (Nelson, 1932), and lobar obstructive emphysema (Sloan, 1953).

Congenital lobar emphysema is of greatest importance and significance in infancy. Cases with or without symptoms are seen from time to time in older children and adults. Furthermore there are occasional mild cases in infancy and Holzel et alii (1956) have drawn attention to this.

Three clinical groups can therefore be recognized:

- Group 1: Congenital lobar emphysema in infancy.
- Group 2: Congenital lobar emphysema, with symptoms, in older children.
- Group 3: Congenital lobar emphysema without symptoms.

The second group can be exemplified by referring to the case of a 6-year-old boy whose presenting symptom was deformity of the chest. X-ray of his chest revealed emphysema of the right upper lobe and the bronchogram showed poor filling of the upper lobe bronchi with crowding and collapse of the middle and lower lobes. Lobectomy was performed and examination of the resected specimen revealed a definite cartilage deficiency. In another case belonging to this group emphysema of the left upper lobe was present (Fig. III). This patient, aged 7, had a history of chronic cough and wheezing and a bronchogram (Fig. IV) revealed compression of all basal bronchi of the left side with lack of filling of the consolidated lingula. Of the rest of the left upper lobe bronchi, only the pectoral was filled but its major bronchi appeared narrowed. The rest of the left upper lobe bronchi showed complete lack of filling.

Cases belonging to the third group, i.e. without symptoms, are reported from time to time. It is interesting to recall the description in 1897 by Wethered, of regional emphysema of the right upper lobe discovered during necropsy on a 51-year-old male and also to draw attention to the case report of Royes in 1938. In his case emphysema of the right middle lobe was found during a necropsy on a man of 31 killed in a motor cycle accident.

The final aspect worthy of discussion concerns the diagnosis and differential diagnosis of the condition. Although cases with chronic or recurrent respiratory infections in later childhood, with lobar emphysema, do occur, their condition is rarely urgent; the situation in infancy may be vastly different and it is important to recognize that breathlessness in babies may be, and often is, due to a condition capable of being ameliorated by surgical means. The significance of

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FIG. III. X-ray of chest showing emphysema of the left upper lobe but excluding the lingular segment.

breathlessness in the first few months of life must be stressed; many "medical" conditions such as respiratory infection, pulmonary agenesis and certain types of cardiac failure may be the basis for this symptom. It is even more important to realize that "surgical" conditions can cause breathlessness. Congenital lobar emphysema is one such condition. Others which enter into the differential diagnosis include congenital diaphragmatic hernia, vascular ring, pneumothorax and pyopneumothorax and lung cysts, congenital and acquired.

There is little place for thoracentesis in the management of congenital lobar emphysema. In fact needling may be dangerous because at the same time as being ineffectual it may lead to the development of a complicating tension pneumothorax. As pointed out on a previous occasion, a problem sometimes occurs when the dual conditions of congenital lobar emphysema and congenital cardiac disease co-exist; under such circumstances if it is not possible to deal with both lesions at the one operation then lobectomy should probably be performed first.

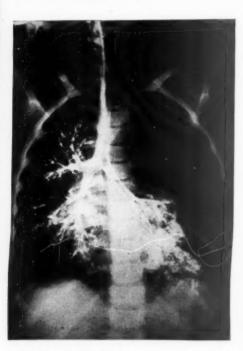


FIG. IV. Bronchogram. (Same case as Fig. III.)

#### SUMMARY

- A case of congenital lobar emphysema is described.
- Attention is drawn to the fact that three clinical groups of this condition can be recognized.
- Although a common aetiological factor has not yet been described, a cartilage deficiency in the bronchi has been demonstrated in many of the reported cases.
- 4. The possible surgical significance of breathlessness in babies is stressed.

#### ACKNOWLEDGEMENTS

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It is a pleasure to record my thanks to Mr. Russell Howard for his generosity in allowing me to study and quote two cases of his and also for his help in the preparation of this paper. The case reported was referred to me by Dr. V. L. Collins, of the Royal Children's Hospital, Melbourne. Photographic assistance was provided by Mr. Murphy of the Department of Photography, Royal Children's Hospital.

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# THE EFFECT OF CORTISONE ON NORMAL AND FRACTURED BONE IN THE RAT\*

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BONE changes associated with cortisone administration have been incompletely investigated and experimental studies have not shown a consistent pattern of skeletal In man occasional spontaneous change. fractures have been reported during cortisone administration (Demartini, Grokoest and Ragan, 1952; Curtiss, Clark and Herndon, 1954; Reifenstein, 1958). Sissons and Hadfield (1955) and Storey (1957, 1958) recorded bone rarefaction in the rabbit though Follis (1951b) found no such change. Birds also develop osteoporosis (Urist, 1958) but cortisone administration has not affected bones of either the guinea pig or mouse (Follis, 1951b). Even in the rat, doses sufficiently high to retard longitudinal bone growth (Sissons and Hadfield, 1955) and inhibit repair of connective tissue (Hurley, Storey and Ham, 1958) do not consistently affect the density of bone; however, with very high doses, 40-50 mg./kg./day of cortisone, dense metaphyseal bone forms due to a reduction in the rate of "osteolytic sequences" (Follis, 1951a). With comparable doses Sobel and Marmorston (1954) found that cortisone had little effect on collagen or bone mineral and, although the hexosamine content of rat femur was reduced, osteoporosis failed to develop.

Healing fractures are sensitive indicators of factors modifying bone growth. Ragan (1950) and Sissons and Hadfield (1951) found that cortisone inhibited repair of fractures in the rabbit but no effect was observed in the rat at a dose level of 20 mg./kg./day (Key, Odell and Taylor, 1952). However, this amount of cortisone per day is of the order of half that required to produce an increase in density of metaphyseal bone in this animal. Using a similar dose level in rats Duthie and Barker (1955) found that the differentiation of cells in callus, up to the formation of chondrocytes, proceeded normally

until the 7th day, but the cartilage mass failed to mature and increased amounts of chondroitin sulphuric acid were liberated with subsequent failure of endochondral ossification. With dose levels of the order of 40 mg./kg./day, Kowalewski (1958) showed that uptake of S35 by callus of fractured rat femur was decreased.

The significance of the above findings is not immediately apparent and histological study of fracture repair in rats at dose levels of cortisone known to affect bone might be expected to elucidate the interrelationships of these observations. Accordingly this first report deals with an attempt to find some consistent pattern in normal and fractured bone during cortisone administration.

#### MATERIALS AND METHODS

The effect of cortisone was investigated in 74 rats (Sprague-Dawley and Wistar) fed Barastoc pellets supplemented by green vegetables and water *ad libitum*. Barastoc pellets are a local proprietary animal food used largely for poultry and contain a high level of Ca and P.

Cortisone acetate was administered at a dose level of 40 mg./kg./day by subcutaneous injection into the inner side of the thigh.

The tibia was fractured, the intact fibula providing partial support for the leg as previously employed by Storey and Varasdi (1958).

Rats were killed at intervals during the experiments; the tibia, femur, skull and mandible were dissected from soft tissue and, in the case of the specimens removed during the early stages of fracture repair, enough muscle was left on the bones to hold them in position until fixed.

Bones were fixed in neutral 10 per cent. formol saline, decalcified in 5 per cent. nitric acid and embedded in paraffin wax. Sections

<sup>\*</sup>Received for publication 8th October, 1959.

were cut at 5 to 7  $\mu$  and the following stains used: Weigert's or Ehrlich's haematoxylin and eosin, Alcian blue, periodic acid Schiff (PAS), silver impregnation and Schmorl's canaliculi stain.

#### EXPERIMENTS

The effect of different dose levels of cortisone—0, 5, 10, 20 and 40 mg./kg./day
—for fourteen days was observed in 10 rats weighing 90-110 g.

In fourteen days control animals doubled their original weight but with increasing levels of cortisone rats gained less until with 40 mg.-kg./day they only increased by 19 per cent.

Macroscopic examination showed that the tibiae grew less and the epiphyseal cartilage became narrower with increasing dose of cortisone. The metaphysis was shorter than normal at 10 mg./kg./day and denser and tougher at 40 mg./kg./day (Figs. I and II).

Microscopic examination showed gradations in change with increasing dose levels from normal to dense metaphyseal bone; the difference between normal and cortisone treated bone is described below.

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The epiphyseal cartilage consists of long columns of cells arranged longitudinally in lacunae separated by intercellular matrix; for purposes of description it can be divided into three zones, proliferating, maturing and calcifying. In the proliferative zone cells are small, spindle shaped and embedded in deeply Alcian blue stain material which accumulates with progressive enlargement of the cell. Alcian blue-staining material then becomes less abundant and gradually disappears with hypertrophy of the cell until only the lacunar capsule and cellular remnants stain. In the matrix which separates the columns of cells in the proliferative and maturing zones, PAS and silver stains show widely-spaced longitudinally arranged fibrils in homogeneous matrix staining irregularly With the continued enwith Alcian blue. largement of cartilage cells in the zones of maturation and calcification these fibrils converge so that although the matrix between the

columns of cells is reduced in width it stains more deeply with PAS but not with Alcian blue (Fig. III).

Cartilage cells in the zone of resorption are replaced by ingrowing vessels leaving thin scalloped remnants of cartilage which stain deeply with PAS and Alcian blue. On the margins of this cartilage osteoblasts containing PAS-staining cytoplasmic granules are associated with the laying down of bone matrix which stains faintly with PAS, not at all with Alcian blue, and contains agyrophilic fibres. In the central area of the metaphysis the bone trabeculae are resorbed while those on the periphery are continuous with and form the diaphysis; here remodelling of lamellar bone occurs to form the shaft.

# (B) Cortisone treated animals

The epiphyseal cartilage is narrowed; the number of cells in columns is reduced particularly in the maturing zone. Chondrocytes in the maturation zone are smaller than normal and few enlarge to the size of those in the control animal. Extracellular Alcian blue-staining material in lacunae walls is still present although reduced in amount. matrix between columns of cells is not decreased in area and PAS-staining fibrils are not completely compressed together even in the zone of calcification. Instead of orderly replacement of hypertrophied cells by ingrowing vessels there is irregular removal of cells and matrix so that wider cartilage cores staining faintly with PAS and Alcian blue remain in the metaphyseal trabeculae (Fig. IV). Some cartilage remnants in the metaphysis contain chondrocytes still in the stage of early maturation.

In the metaphysis, material staining deeply with PAS and haematoxylin, is laid down on cartilage matrix margins followed by lamellar bone so that eventually cartilage cores are encased by bone. In the remodelling zone osteoblasts are small, spindle shaped and stain deeply with haematoxylin while resorption is reduced and bone in the central area is not removed. Further remodelling of trabeculae in the diaphysis does not occur so the metaphysis ends abruptly at the diaphysis. Here litle sign of lamellar bone formation is seen and bone margins are lined by either small spindle shaped or occasional multinucleated cells.

Sections of skull show little change except inhibition of appositional and sutural bone growth. At the bony margins of the suture, instead of the normal osteoid border, an irregular line staining darkly with haematoxylin or PAS is present and osteoblasts, small and spindle shaped, are few in number.

(2) The effect of cortisone on rats of different ages was tested in three groups each of twelve rats weighing 20-28 g. (suckling), 90-110 g. (growing) and 300-310 g. (adult) for twenty-five to seventy days.

Cortisone treated rats in the youngest age group gained weight; slowly growing rats either failed to gain or lost little while adults lost an average of 15 per cent. in weight in seventy days.

Macroscopic examination of cortisone treated rats showed a wide dense metaphysis in suckling rats, a narrow dense one in growing rats and little change in adult rats. However histological sections show that even in adult animals metaphyseal trabeculae are denser and enclose wide cores of unresorbed cartilage similar to those seen in cortisone treated rats in younger age groups (Figs. V and VI).

(3) The effect of cortisone on the repair of fracture of the tibia was tested in 28 rats weighing 90-110 g. The animals were divided into two groups, one treated with 40 mg./kg./day of cortisone, the other acting as control; rats were killed at four, six, eight, twelve, sixteen, twenty and twenty-four days after fracture of the tibia.

#### (A) Control animals

#### Macroscopic examination

In the control group the callus reached its maximum size at twelve days and on section the middle and outer areas consisted of cartilagenous material, the remainder of bone; from this stage onwards there was a progressive decrease in cartilage and more bone appeared until by 24 days bony union had occurred and most of the cartilage had been removed. By this time the continuity of the

marrow cavity had been partly restored by removal of internal callus (Fig. VII).

Microscopic examination

By four days a callus forms around the ends of the fractured bone. Silver impregnation shows that fibrils run from the elevated periosteum into the ends of the newly forming bone trabeculae to form an orientated woven fibrous matrix.

Within and at the margins of this newly forming bone are plump osteoblasts containing PAS-staining cytoplasmic granules. In contrast with old bone this new matrix stains deeply with PAS due to the accumulation of fine granular material but fails to stain with Alcian blue except for the trabecular margins where a thin lightly staining osteoid border is present. In the cartilagenous part of the callus the chondrocytes contain some cytoplasmic PAS-staining granules, while the matrix stains intensely with Alcian blue but not with PAS. Agyrophilic fibres in the periphery of the callus connect the ends of the fractured bone and in the central area removal of haematoma and debris occurs by ingrowing granulation tissue. The fractured ends of the original bone are now honeycombed and vascularity in the area is increased.

By eight days the callus is larger, periosteal bone trabeculae are longer and orientated at an angle of 60-70 degrees to the bone shaft and more cartilage has formed. Replacement of cartilagenous callus by bone is now proceeding by the usual process seen during endochondral ossification, although less orderly than in the metaphysis of long bones (Fig. VIII).

By the twelfth day, when the callus reaches its maximum size, union of the two halves of the fractured bone is by connective tissue fibres. Resorption of the newly formed bone trabeculae of the callus is now in progress and large areas previously occupied by orientated bone or bone formed from cartilage are filled with dilated and more numerous vessels. A new and more mature lamellar form of bone begins to form which stains less intensely with PAS and fails to stain with Alcian blue; silver impregnation demonstrates the dense lamellar nature of the matrix..

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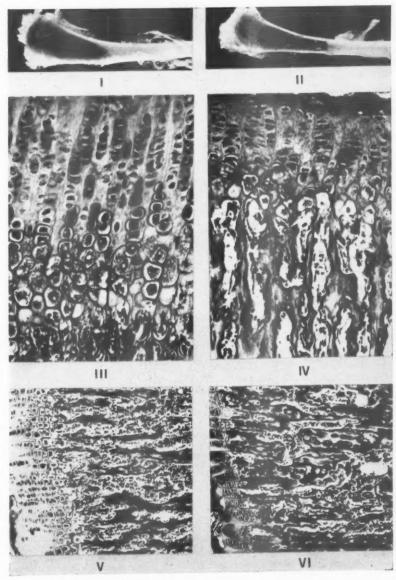


FIG. I. Tibia of normal young rat.

FIG. II. Tibia after fourteen days cortisone treatment showing decrease in size, narrow epiphyseal cartilage and dense metaphysis

FIG. III. Photomicrograph of normal epiphyseal cartilage showing deeply staining Alcian blue material around cartilage cells in zones of proliferation and maturation. Smaller amounts are present in the fibrous matrix between columns of cells. Alcian blue, cosin. (x 150)

FIG. IV. Photomicrograph of narrow epiphyseal cartilage after fourteen days cortisone administration. The maturation zone is almost absent and Alcian blue-staining material is decreased around cells, but is still present in fibrous matrix. Alcian blue, cosin. (x 150)

FIG. V. Photomicrograph of metaphyseal area of an old rat. Weigert's haematoxylin and cosin. (x 100)

FIG. VI. Photomicrograph of metaphyseal area of an old rat treated with cortisone for twenty-eight days. The epiphyseal cartilage is narrow and wide metaphyseal trabeculae contain large cores of cartilage matrix. Weigert's haematoxylin and cosin. (x 100)

By twenty to twenty-four days the picture is that of a fracture partly healed by bony union with only remnants of cartilage and connective tissue occurring between the bone ends. Remodelling processes continue in the callus with the removal of immature bone and cartilage and its replacement with mature bone in the external callus; gradual removal of trabeculae from the internal callus restores the continuity of the marrow cavity (Figs. X, XII and XIV).

# (B) Cortisone Treated Animals

Macroscopic examination

The callus was slightly smaller than normal and contained more cartilagenous material than bone at the end of twelve days. After twenty-four days the callus was harder to cut and appeared denser than control bone but still contained cartilage separating the fractured ends of the leg. The metaphysis was also denser than normal (Fig. VII).

# Microscopic examination

After four days the callus is slightly smaller and contains relatively more cartilage than bone; silver impregnation shows that bone matrix consists of woven fibrils. At eight days, periosteal bone trabeculae are shorter and less uniformly orientated than those in the control callus. In cartilage, chondrocytes are smaller and the area occupied by ground substance staining irregularly with Alcian blue is larger; endochondral ossification has not yet begun (Fig. IX).

At twelve days more cartilage remains in the external callus and although endochondral ossification has begun it proceeds in a much less orderly manner than in the control fracture. In the zone of resorption chondrocytes have not enlarged normally so that relatively more intercellular matrix remains. This stains irregularly with Alcian blue and lightly with PAS.

Resorption of cartilage is irregular and wide cartilage remnants still containing maturing chondrocytes occur in the bony callus. Here trabeculae are denser and osteoblasts at their margins are smaller, more darkly staining and contain fewer PAS-staining granules than normal. The ends of the fracture are connected peripherally by connective tissue fibres but in the central area large remnants of haematoma still remain.

At sixteen days, endochondral ossification is appreciably less than normal and large areas of cartilage still remain in the callus. Throughout the cartilage chondrocytes are small and many now fail to stain. Trabeculae in the bony callus are thick and contain enclosed wide cores of cartilage matrix. Little remodelling of immature bone has occurred, although a few small resorption cavities are present.

By twenty to twenty-four days, although endochondral ossification continues, much cartilage remains between the two ends of the fractured bone, while the bony callus still consists largely of dense trabeculae either enclosing immature unresorbed bone or unresorbed calcified cartilage (Figs. XI, XIII and XV). Resorption of the shaft of the fractured bone has not progressed as in the normal animal and bone trabeculae still persist in the marrow cavity. In some areas small circumscribed layers of bone appear which are separated from the immature bone by a deeply haematoxylin and PAS-staining reversal line. These layers of bone are arranged in small, but distorted, Haversian systems and appear only in the external callus (Fig. XV). Silver impregnation shows their matrix to consist of dense lamellar bundles of fibres which stains less intensely with PAS than woven bone. Cells at the trabecular margins are small and spindle; small multinucleated osteoclasts lie in Howship's lacunae. Osteoclasts are considerably fewer than in control animals.

There is a considerable variation in the formation of lamellar bone in the callus. Animals which fail to gain, or which lose, weight develop little mature bone while rats with only slightly reduced weight gain form larger amounts of lamellar bone.

The fracture ends are joined on the posterior (concave) aspect by cartilage and on the anterior (convex) aspect by a wide band of connective tissue in which small islands of cartilage still remain. In the marrow space at the site of fracture is a mass of connective tissue occupying the area of the haematoma and containing a few small wandering cells and multinucleated cells lying some distance from the nearest bone.

VII VIII

FIG. VII. Photograph of tibiae from normal (above) and cortisone treated (below) rats twenty-four days after fracture. The callus in the cortisone treated rat is denser than normal.

FIG. VIII. Photomicrograph of posterior aspect of callus at eight days in a normal rat showing new woven bone trabeculae orientated at an angle to the original bone with abrupt transformation to cartilage. Ehrlich's haematoxylin and cosin. (x 50)

FIG. IX. Photomicrograph of posterior part of a callus at eight days in a cortisone treated rat showing slightly decreased bone and cartilage formation. Haematoxylin and cosin. (x 50)

FIG. X. Photomicrograph showing endochondral ossification in callus of a fractured tibia after twenty days. Haematoxylin and cosin. (x 50)

FIG. XI. Photomicrograph showing endochondral ossification in a cortisone treated rat after twenty days. Bone trabeculae are wider than normal. Haematoxylin and cosin. (x 50)

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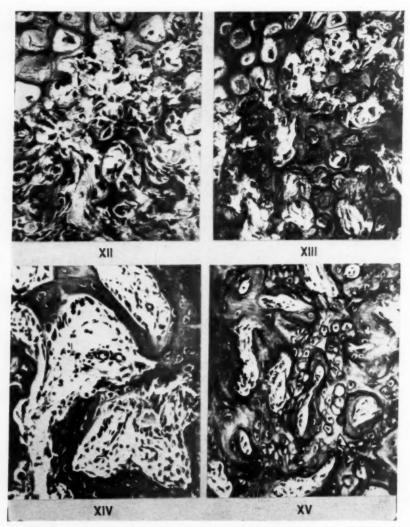


FIG. XII. Photomicrograph showing endochondral ossification in a callus of a fractured tibia after twenty-four days. Bone is forming on thin cartilage remnants which stain deeply PAS. (x 475)

FIG. XIII. Photomicrograph showing endochondral ossification in a callus of a fractured tibia after twenty-four days cortisone treatment. Cartilage cells and lacunae are smaller and relatively more intercellular material remains unresorbed compared with the control. PAS. (x 475)

FIG. XIV. Photomicrograph of bone in the callus of a normal rat at twenty-four days. Extensive remodelling of bone is in progress. Haematoxylin and eosin.  $(x\ 200)$ 

FIG. XV. Photomicrograph of bone in the callus of a cortisone treated rat at twenty-four days. Large amounts of woven bone remain unresorbed and a deeply staining reversal line marks the junction between immature and lamellar bone. Haematoxylin and cosin. (x 200)

#### DISCUSSION

When cortisone at a dose level of 40 mg./kg./day is administered to rats they fail to grow normally and dense bone develops in the metaphysis of long bones. Decreased rate of growth is one characteristic feature of the action of cortisone and in bone is shown by narrowing of the epiphyseal cartilage and cessation of appositional and sutural growth. In addition the metaphysis of long bones becomes denser largely due to failure of resorptive processes so that wide dense cores of unresorbed cartilage matrix enclosed by lamellar bone remain in the metaphyseal region. Similar changes occur during endochondral ossification in the callus of fractures in cortisone-treated rats. However, despite general growth inhibition, the formation of the callus in the early stages is only slightly retarded. These findings are not in agreement with those of Key et alii (1952) who found little difference in the healing process when using a dose of 20 mg./kg./day of cortisone. With this same dose Duthie and Barker (1955) found little delay in cartilage formation during the proliferative phase but, following this, there was failure of endochondral ossification in the callus, which calcified rather than ossified. Different results of various investigators can be attributed partly to the use of different dose levels of cortisone and partly to experimental methods; for instance Key et alii (1952), in contrast with other workers, inserted pins into the marrow of fractured bones in order to immobilize them. Also, no consistent change has been observed in rat bone on a dose of 20 mg./kg./day of cortisone (Winter, Silber and Stoerk, 1950; Follis, 1951a; Sissons and Hadfield, 1955).

One consistent difference between the effect of cortisone on normal and fractured bone is in the formation of cartilage. At endochondral growth sites proliferation of cells and formation of cartilage is slowed whereas almost normal amounts can form in an early callus. This may be the result of a difference in the degree and type of stimulus; for example, with fractures of bones mechanical stress would be high whereas in growing endochondral cartilage it would be considerably less. Furthermore, the degree of oedema induced by different stimuli varies greatly

with cortisone; the oedema of thermal burns is unaffected while that of the Arthus reaction is greatly suppressed (Spector, 1958).

The pattern of change in both normal and fractured bones following the proliferative phase in rats treated with cortisone is similar and is associated with a delay in the rate of bone remodelling. Thus the present experiments show that both formative and resorptive processes are affected by cortisone. In growing cartilage maturation of chondrocytes is delayed, cells do not enlarge normally so that the amount of intercellular fibrillar matrix remaining at the zone of resorption is proportionately greater than normal. In the zone of maturation extracellular Alcian bluestaining material, presumed to be ground substance (Menzies and Mills, 1957), is decreased in lacunae walls in contrast with control animals. This delay in maturation of cells with appearance of relatively more cartilage matrix may possibly explain Duthie and Barker's (1955) finding of increased liberation of chondroitin sulphuric acid in cartilage matrix in the callus of cortisone treated rats.

Subsequent to uneven maturation, diminished and irregular resorption leaves wider cores of cartilage matrix around which lamellar bone develops to form dense metaphyseal trabeculae; resorption of this bone is also delayed so that a sclerotic metaphysis remains. Follis (1951a) was of the opinion that this dense metaphyseal bone resulted from a retardation of normal "osteolytic sequences" while Duthie and Barker (1955) thought that cortisone affected the "formation of the periosteal cartilage blastema and subsequent process of endochondral ossification." The present experiments show that these views are not inconsistent as, in addition to failure of chondrocyte enlargement which leaves more cartilage matrix to be removed, there is overall reduction of bone resorption at every stage of the remodelling process.

The findings of decreased S<sup>35</sup> uptake in healing fractures (Kowalewski, 1958) and decrease in hexosamine in the bone (Sobel and Marmorston, 1954) of cortisone treated rats may be related both to a decrease in production or change in the nature of Alcian blue-staining mucopolysaccharide associated with delayed maturation and final smaller

size of cartilage cells. Sobel and Marmorston (1954) suggest that decrease of hexosamine containing mucopolysaccharide may be a necessary prerequisite for the development of osteoporosis. However reduction in ground substance is no indication of and may not be directly related to the development of osteoporosis. For osteoporosis to develop, in addition to abnormality of bone matrix formation, resorption of bone must proceed (Storey, 1957). Indeed in the present experiments where resorption is delayed, metaphyseal bone becomes sclerotic instead of porotic.

In contrast with the changes seen in the rat, rabbit bone rapidly becomes porotic during cortisone treatment. This rarefaction has been attributed to an anti-anabolic action in the presence of continuation of normal (Sissons and Hadfield, 1955) or increased bone resorption (Storey, 1957, 1958). As with the rat, this difference may be one not of interpretation of histological findings, but of differences in dose of cortisone used in the rabbit.

Thus, given a similar degree of inhibition of growth, the rate of resorption could well be a major factor determining the nature of the bone changes and is largely responsible for the species difference between cortisone treated rabbits and rats.

#### SUMMARY

Cortisone treated rats develop dense metaphyseal bone and, during the healing of fractures, dense callus. At endochondral growth sites there is delay in maturation of chondrocytes and decreased extracellular Alcian blue-staining mucopolysaccharide. tion of cartilage is decreased so that more remains encased by bone in the metaphysis where trabeculae in their turn are not remodelled to form the diaphysis. In contrast to epiphyseal cartilage early callus growth is not greatly inhibited, but similar changes occur during remodelling. The result is a dense callus consisting of bone in various stages of maturation, prolonged retention of cartilage and failure of bony union. Thus in

contrast to rabbits, where increased resorption is associated with osteoporosis, rats develop dense bone due partly to decreased resorption.

#### ACKNOWLEDGEMENT

This work was carried out under a grant from the National Health and Medical Research Council of Australia. The cortisone acetate was supplied by Boots Pure Drug Co. (Australia) Pty. Ltd., to whom we extend our sincere thanks.

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# THE PRINCIPLES OF STEREOTACTIC BRAIN SURGERY AND ITS USE FOR THE RELIEF OF INTRACTABLE PAIN\*

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STEREOTACTIC brain surgery may be defined as the technique of aiming an instrument at a target within the cranial cavity with the aid of predetermined mathematical co-ordinates. It enables the surgeon to stimulate or destroy tissue at a depth without damage to superficial structures.

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Guiding instruments for this purpose have been used by experimental physiologists for more than eighty years. Spiegel and Wycis (1952) refer to the experimental destruction of the vasomotor centre in the medulla of rabbits reported by Dittmar in 1873. Horsley and Clarke in 1908 developed an accurate stereotactic apparatus which, with minor modifications, is still used for animal experiments today. For more than seventy years the method remained confined to experimental neuro-physiology. Spiegel and Wycis (1947) of Philadelphia were the first to report the application of stereotactic surgery for operations on the human brain. This delay in the adoption of stereotactic methods for therapeutic purposes appears to be due mainly to the variability in the size and shape of the human skull and the lack of a consistent relationship between cranial landmarks and intracerebral structures. As an example of this, it was found that the position of the pineal gland in an antero-posterior direction varies by as much as 15 mm., when related to a plane constructed through the centre of the external auditory meati. In the cat, on the other hand, the size and shape of the skull and the cerebro-cranial relationships are remarkably constant.

Progress in the knowledge of the anatomy and physiology of the nervous system during the last fifty years provided the essential foundation for surgical attempts at relief of pathological conditions by the selective destruction of specific nuclei or fibre tracts. The last ten years have seen the progressive improvement of stereotactic methods and a

variety of instruments are now available for the use of surgeons with routine hospital facilities.

As stereotactic brain surgery involves the destruction of brain tissue, the selection of a technique and instrument must depend on the following criteria:

- Technical perfection and accuracy of the instrument for anatomically exact placement of the lesion.
- 2. Safety and comfort to the patient and freedom from possible complications.
- Adoption of a standard procedure which permits a critical analysis and comparison of results.

The wide variation in the relationship between human intracerebral landmarks and in the size of subcortical nuclei modified by disease makes it impossible to fulfil these demands completely. The technical design of the instrument, however, must provide for an accuracy in the range of 0.5 mm. to guide a cannula or electrode exactly at the chosen target.

Several instruments have been developed in various parts of the world and each probably has its own virtues and advantages. We have chosen an instrument designed by Riechert and Wolff (1951) of Freiburg, Germany and later modified by Riechert and Mundinger. After using it for more than fifteen months in over 120 operations, we consider that it provides the necessary accuracy and safety for this form of surgery.

The majority of our operations were done for the relief of Parkinsonism and the results will be reported in a separate paper. The safety of our method can be demonstrated by the fact that no immediate post-operative death nor permanent hemiplegia occurred in our series and that only one patient suffered a post-operative haemorrhage into the midbrain and died five months after operation.

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The variation in the anatomy of the human brain, accentuated by pathological processes, constitutes the main problem. The measured distances between intracerebral landmarks, and the angle formed by the axis of the brain stem with the cerebral hemispheres vary considerably from patient to patient. The wide variation in the position of the pineal gland was mentioned earlier and anatomical studies have shown that similar deviations from the average apply to the distances between other radiologically visible landmarks, as well as to the length of the thalamus and other parts of the basal ganglia. The relative position of subcortical nuclei may be altered by cerebral atrophy, enlargement of the ventricles and other pathological processes, and this variation in size is not necessarily the same in the sagittal and coronal dimensions.

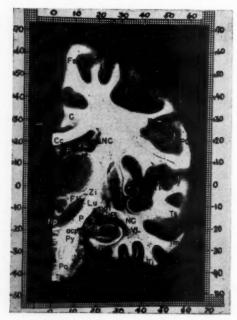


FIG. I. Coronal macroscopic section of the human brain at a level 15 mm. anterior to the pineal gland. (Reproduced from Spiegel and Wycis, 1952.)

Several atlases of coronal and sagittal sections of the brain at various levels are available and contain measurements averaged from series of brains of patients of varying age. We have relied on the atlas published by Spiegel and Wycis (1952) which includes variability studies of the distances of several

subcortical nuclei from radiologically visible landmarks.

Fig. I, reproduced from this atlas, shows a coronal section 15 mm. anterior to the pineal gland and indicates the close anatomical relationships in the region of the thalamus and basal ganglia and the need for absolute accuracy in localisation. The scale at the margin of this figure is in millimetres.

As the subcortical structures which are the target of these operations bear no constant relationship to bony landmarks of the skull, the position of the intended lesion must be determined in relation to the ventricular system. Measurements obtainable from the atlas provide the average distances of various subcortical nuclei from specific landmarks such as the foramen of Monro, posterior commissure or septum pellucidum. An air encephalogram with good filling of the ventricular system, showing these landmarks, is therefore an

Basic principles of stereotactic brain surgery

essential part of the operation.

To permit an accurate localization of intracerebral structures, the measurements of our individual patient's brain must be compared with the average measurements of the atlas in three dimensions and a relation factor between the two brains is calculated for each dimension. All further measurements for the subcortical target in our patient must be corrected by these relation factors. The average distance of the target from radiologically visible landmarks is determined from the atlas, corrected by the relation factors, and marked on X-ray films in antero-posterior and lateral projections. The position of the target has now been established in three dimensions in relation to the sagittal, coronal and basal planes of the skull.

The accurate aiming of an instrument towards this target is difficult because of the irregular shape of the human skull and its contained brain. This geometrical problem can be solved by surrounding the skull by an exact sphere and relating the target of the planned lesion to a sagittal, coronal and basal plane, each passing through the centre of this sphere. These three planes must be at right angles to each other, and they provide the three co-ordinates in relation to which all measurements are taken.

A metal ring surrounding the patient's skull represents the circumference of such a sphere in the basal plane, and on this a vertical sagittal plane passing from 0° to 180° and a vertical coronal plane extending from 90° to 270°, each passing through the centre of the sphere, can be erected. The site of the target can now be determined accurately in three dimensions by its distance from each of these three planes.

In the diagram (Fig. II) the metal ring forming the circumference of a sphere in the basal plane is shown as ACBD. Vertical planes are erected on the diameter between A and B, and at right angles to this on the diameter between C and D, while the horizontal plane of the ring itself establishes the third or basal dimension, which also passes through the centre of the sphere.

N represents the target aimed for within this sphere. It can be determined by measurement of its distance from the vertical sagittal plane, XR, from the vertical coronal plane, YR, and of its level above or below the horizontal basal plane, RN.

Operative technique

The operation may be divided into four stages:

 Radiological localization of the site of the intended lesion.

After premedication with pentobarbitone, chlorpromazine and atropine and under light pentothal anaesthesia, a lightweight metal ring made of an aluminium alloy and marked in degrees from 0° to 360° is attached to the patient's head. The ring is firmly fixed into position by six pins screwed into the outer table of the skull. Local anaesthetic is infiltrated into the scalp at the site of perforation of the pins. Fig. III shows the ring attached to the patient's head.

With the patient sitting up, 120 cc. of air are introduced into the spinal canal by lumbar puncture and a sitting-up X-ray film is taken to show the posterior end of the third ventricle, aquaeduct and cisterna ambiens. The patient is then laid down and the ring is fixed into a clamp so that it stands exactly vertical to the X-ray casette. The position of the ring is further adjusted until the coronal plane, passing through the diameter from 90° to 270°, is truly horizontal and parallel with the central beam of the X-ray.

A metal rod, 10 cm. long, and divided by discs at a distance of 1 cm. from each other, is strapped on to the patient's skull near the midline. It is used for the calculation of the distortion of the X-ray film.

Antero-posterior and lateral films of the encephalogram are then taken. Sighting rods

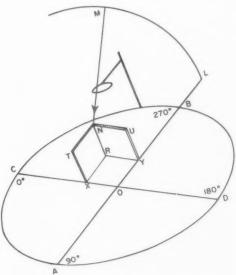


FIG.II. Diagram in which N represents the site of the lesion aimed for, MN is the electrode passing through the burr hole or marking ring, and LM is portion of the arc to which the electrode carrier is attached.

with rings at their tip are attached to the base ring at opposite ends in the coronal and sagittal planes, and a special sighting lamp is fixed to the side of the X-ray tube. When the beam from this lamp passes through the sighting rings and their shadows are superimposed, the central beam of the X-ray will travel exactly through the coronal or sagittal diameter of the ring. Sighting pins, corrected in size for X-ray distortion, are screwed into the base ring at 0° and 180° for the anteroposterior projection and at 90° and 270° for the lateral projection, and must be exactly superimposed on the films if these are correctly taken.

On the X-ray films (Figs. IV and V), the superimposition of the pins is well shown. The 10 cm. distortion rod is seen in the lateral

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projection. Its length on the X-ray is measured and related to its actual length, providing an X-ray distortion factor by which all measurements made on the lateral X-ray film must be corrected. A similar correction factor must be determined for the anteroposterior projection.

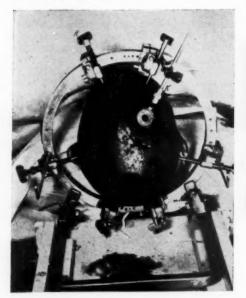


FIG. III. The ring, attached to the patient's skull, is supported in clamps. The burr hole marking ring, and two X-ray sighting rods are shown.

The next step is the comparison of the size of our patient's brain in three planes with the average measurements available from the atlas. For this we use the distance between the posterior border of the foramen of Monro and the posterior commissure in the sagittal plane, the dimension from the septum pellucidum to the upper and outer angle of the lateral ventricle in the coronal plane, and the distance from the foramen of Monro to the dorsum sellae in the vertical plane.

For example, if the X-ray distortion factor is 10/12, and the distance between the foramen of Monro and the posterior commissure on the X-ray is 28 mm., then the true distance between these structures in our patient's brain will be  $\frac{28 \times 10}{12} = 23.3$  mm. The average distance between the foramen of Monro and the posterior commissure has been determined

from the atlas as 25 mm. Hence our patient's brain in the sagittal dimension of the basal ganglia is smaller than average, namely  $\frac{23.3}{25}$  = 0.93, and this correction factor has to be applied to all future measurements in this plane. The same principles apply to the deter-

mination of correction factors in the other



FIG. IV. Lateral view of air encephalogram with the ring in position, showing the superimposed sighting pins, and the X-ray distortion rod. The foramen of Monro and posterior commissure are well seen.

Assuming that we have selected the posterior ventral nucleus of the thalamus as the site of the lesion in our patient, the average distance of the centre of this nucleus in three planes, in relation to the posterior commissure and to the septum pellucidum can be determined from the atlas. This average distance has to be corrected in each plane for the size of our individual patient's brain. In this example the posterior ventral nucleus lies 3 mm. in front of the posterior commissure, and this would be corrected in our patient to  $3 \times 0.93 = 2.8 \text{ mm}$ . To mark this point on the lateral X-ray film, it must again be enlarged by the X-ray distortion factor, i.e.  $\frac{2.8 \times 12}{2.8 \times 12} = 3.36$  mm. Similar measurements

and corrections are made for the distance of the nucleus above the posterior commissure and lateral to the septum pellucidum, and the points so determined are marked on the lateral and antero-posterior X-ray films.

Relation of the site of the intended lesion to the three co-ordinates passing through the centre of the sphere.

It was mentioned above that the central beam of the X-ray must pass exactly along the centre of the basal plane of the ring surrounding the patient's skull, through the coronal plane in the lateral view, and in the sagittal plane of the sphere in the anteroposterior view. A horizontal line passing through the centre of the ring and parallel with the ring is drawn on both X-ray films. In the lateral projection, a vertical line at right angles to this is erected through the centreing pins fixed to the ring at 90° and 270°. This establishes the vertical coronal plane through the centre of the sphere. In the antero-posterior view a similar vertical line is drawn at right angles to the ring through the centreing pins at 0° and 180° and represents the vertical sagittal plane through the centre of the sphere.

The point marking the site of the planned lesion on the X-ray films can now be related to each of these three planes of the sphere, but the distances measured from the X-ray films must again be corrected by the radiological distortion factor for each projection.

We have now established the distance of our target in three dimensions from the three co-ordinate planes of the sphere, represented by XO, YO and UY in Fig. II.

 Aiming and introduction of the electrode, and radiological and physiological confirmation of its position.

The electrode is inserted through a burr hole, which can be prepared either at a prior operation, or immediately before attaching the ring to the patient's head.

To determine the direction and depth of the electrode, the ingenious idea of a "mock operation" is used. An exact replica of the base ring attached to the patient's skull, with identical gradations, is employed for this purpose. This so-called "phantom ring" contains two bars at right angles to each other and parallel to the plane of the ring, and a third bar vertical to this plane. Each is graduated in measurements of 0.5 mm., and they can be moved in three planes. The upper end of the vertical bar has a pointed knob, which represents the site of the target within the sphere. Using the distances of the planned lesion from the three co-ordinates of the sphere, determined from the corrected X-rays, the knob on the phantom is set to represent the target. This knob will now be in the same position in relation to the phantom ring as the centre of the posterior ventral nucleus is in relation to the ring surrounding the patient's head.

As the electrode has to pass through a burr hole on the surface of the patient's skull, the position of this burr hole must be marked also on the phantom. This is achieved by a burr hole marking ring, carried on adjustable arms, which is shown in Fig. III attached to the ring surrounding the patient's head and overlying the site of the burr hole. This marking ring on its carrier is then transferred to the identical position on the phantom ring and the place of the burr hole is thereby marked in space.

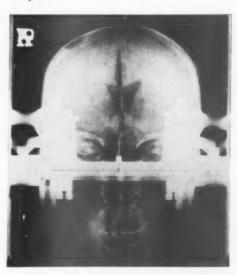


FIG. V. Antero-posterior view of air encephalogram with the ring in position, and sighting pins in the sagittal plane.

A graduated arc with an electrode carrier and guide which can be moved along measured distances in three planes, is attached to the phantom ring. The electrode is then passed through the guide carrier and aimed through the burr hole marking ring, and the carrier is moved about until the tip of the electrode touches the knob representing the target on the phantom (Fig. VI). The electrode carrier is then fixed firmly into place on its arc, and its position in various planes is read from the gradations and recorded. The depth to which the needle penetrates is read from a graduated depth marker attached to the electrode carrier and is also recorded.

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The needle is now withdrawn from the guide and the arc with the electrode carrier is transferred to the ring surrounding the patient's head and is fixed to it in the same position as it was on the phantom. The recorded data of the position of the arc and electrode carrier are checked, and the electrode can now be introduced through the patient's burr hole into the brain to the measured depth and in the direction determined, so that its tip will lie exactly at the target aimed for (Fig. VII).

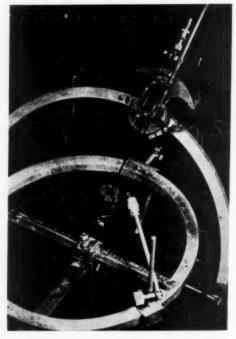


FIG. VI. Phantom ring with arc and electrode carrier. The electrode passes through the burr hole marking ring and its tip is in contact with the knob representing the target.

The position of the electrode is checked by further X-rays in antero-posterior and lateral projections and compared with the point calculated and drawn on the initial films (Figs. VIII and IX).

Physiological control of the correct position of the electrode in the subcortical target is achieved by an electric stimulator, producing 3 c.p.s. waves of up to 40 volts across the circuit. If the electrode tip is in the sensory thalamic nuclei, this stimulation may reproduce pain; if in the lateral ventral nucleus selected as the target in our operations for the relief of Parkinsonism it will reproduce the tremor.

#### 4. Production of the lesion.

Once the correct position of the electrode tip is confirmed by radiological and physiological control, the lesion in the selected subcortical nucleus can be made. Following Riechert's method, we have relied on electrocoagulation to destroy the tissue. The electrodes, excepting their bare tips which are 2 mm. or 3 mm. long, are insulated and are carried and introduced within a fully insulated cannula. A radio frequency current of up to 200 milliamperes from a diathermy machine is used and repeated coagulations, each of about ten seconds, are made in the same spot to avoid over-heating and damage to the insulating material.

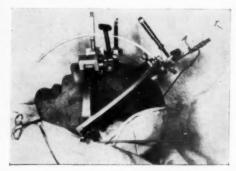


FIG. VII. The arc and electrode carrier are attached to the ring surrounding the patient's skull. The upper end of the electrode and its cannula are seen. The X-ray sighting rods are fixed to the ring in the coronal plane.

A second electrode with a bare tip protruding laterally through a hole on the side of its cannula is employed to enlarge the lesion. Its tip can be extended in any desired direction to a distance measurable from a scale at the top of the cannula, and the size of the lesion can be increased in various directions until a satisfactory result is produced.

During the last stage of the operation the patient is usually fully conscious and able to co-operate. This permits the surgeon to test his cutaneous sensation, motor power, tone and reflexes and to prevent any possible encroachment on the pyramidal tract in the internal capsule.

The variability in the extent of electrolytic lesions was studied by Szekely (1956), who reported a statistical evaluation of the size of 146 anodal electrolytic lesions, produced in cats in the thalamus, amygdala, and midbrain. He employed a direct current of 10 mA flowing for one minute, and an electrode wire protruding a length of 3 mm. The average length of the lesion was  $5.39 \pm 0.68$  mm. and the average width  $2.50 \pm 0.50$  mm.

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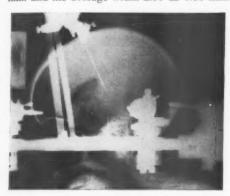


FIG. VIII. Lateral view of air encephalogram showing the electrode in position.

Histologically the electrolytic lesion consists of a central cavity, an intermediate zone of coagulation, and a peripheral zone of oedema and inflammatory reaction. Occasionally small haemorrhages may surround the lesion. In the peripheral zone of oedema nervous impulses are temporarily blocked, but will recover as the oedema subsides after two to three days. This explains why abolished at operation may occasionally recur during the first few post-operative days, if the lesion was made too small. In some of these cases we have enlarged the lesion by a second operation and succeeded in abolishing the tremor completely.

The application of stereotactic brain surgery for the relief of intractable pain

Two years after the introduction of human stereotactic brain surgery, Wycis and Spiegel (1949) reported their results of the selective destruction of the spino-thalamic tracts in the midbrain in 16 patients suffering from intractable pain. They applied the term mesencephalotomy to this operation.

Riechert (1957) reviews the methods and results of stereotactic procedures for the relief of pain reported by Wycis and Spiegel,

Talairach et alii, and Monnier, and describes the results in his own series of 17 patients. The indications for operation in these reported series were intractable pain due to post-herpetic neuralgia (mostly in the ophthalmic division of the trigeminal nerve), painful phantom limbs, thalamic syndromes and pain caused by advanced malignant disease.

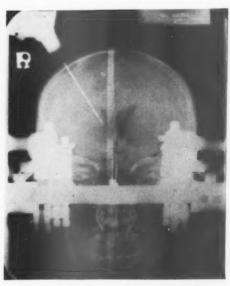


FIG. IX. Antero-posterior view of air encephalogram showing the electrode in position. In this film the central beam of the X-ray does not pass exactly through the sagittal diameter of the ring, and the sighting pins are therefore not superimposed.

While Wycis and Spiegel (1949) chose the spino-thalamic and quinto-thalamic tracts in the midbrain at the level of the superior colliculus as their target, Riechert selected the basal part of the posterior ventral nucleus of the thalamus as the site of his lesions. In this nucleus the body is topographically represented. The sensory fibres from the face end in its medial part, the arcuate nucleus, those from the upper limb and trunk occupy an intermediate position, and the afferent fibres from the leg terminate in its most lateral part.

In 9 of Riechert's 17 patients the pain was either abolished or greatly relieved, in 5 it was only temporarily improved, one was no better, and two were worse after the operation. Talairach et alii (1949) destroyed the pos-

terior ventral nucleus of the thalamus in twelve elderly patients suffering from a thalamic syndrome of vascular aetiology. They were able to relieve the pain in one half of these and two patients died as the result of operation.

In patients where the pain is elaborated by emotional disorders the dorso-medial nuclei of the thalamus on both sides may be destroyed and Wycis and Spiegel (1949) have combined such lesions with a mesencephalotomy in some of their cases. The dorso-medial nucleus of the thalamus projects to the pre-frontal cortex, particularly to the orbital surface of the frontal lobe, and its destruction may result in personality changes similar to those following on pre-frontal leucotomy. It is claimed that undesirable psychological and intellectual changes do not occur as frequently after coagulation of the dorso-medial nuclei as after pre-frontal leucotomy.

Riechert (1957) summarises the results reported in the literature up to that time, and indicates that intractable pain was successfully relieved in about 50 per cent. of all cases. The post-operative mortality of 12 to 16 per cent. reported in some series is not excessive, considering that many patients are of advanced age and poor operative risks.

The risk of hemiparesis from encroachment on the internal capsule, of possible damage to the red nucleus or oculomotor nucleus in deeply placed lesions and the theoretical possibility of producing a thalamic syndrome by the electrolytic lesion must be considered. These complications, however, occurred only in a small proportion of the reported operations.

It must be remembered that the anatomical and pathological variations between individual brains, discussed earlier in this paper, make it impossible to place the lesion with absolute accuracy in every case. This accuracy is much more critical in operations aimed at subcortical pain centres than in those for the relief of Parkinsonism, and the reported incidence of complications is much greater. A precise stereotactic instrument and method are essential and the pseudo-stereotactic procedures used in various centres for the treatment of hyperkinetic disorders cannot be relied on for this indication. Considering the

mortality, the risk of complications and the 50 per cent. chance of success, only patients with severe pain which failed to respond to other therapeutic measures, should be considered for stereotactic brain surgery.

We have operated on 7 patients, 5 males and 2 females, for the relief of pain. Five of these were over 67 years old, and the remaining two were in their forties. One man, aged 71, died five days after operation from an extensive cardiac infarction. No serious or permanent complications occurred in the others. In two patients the pain was completely abolished, two obtained great relief and the remaining two had moderate relief sufficient to enable them to return to an active existence.

In two cases the operation was performed for the relief of severe and intractable neuralgia following on ophthalmic herpes zoster. The lesion was placed in the medial part of the posterior ventral nucleus of the thalamus (arcuate nucleus). In both the pain was abolished in the face, but moderate pain in the affected eye persisted. One of these patients died from a cardiac infarction on the fifth day after operation and autopsy confirmed the centre of the electrolytic lesion in the arcuate nucleus. A small haemorrhage surrounding the lesion extended into the lateral part of the posterior ventral nucleus of the thalamus (Fig. X) (Cases 1 and 2 in the appendix).

One patient suffered from post-herpetic neuralgia involving the skin of the left side of his chest. He obtained great relief after coagulation of the lateral part of the posterior ventral nucleus of the right thalamus. Slight pain in the affected dermatome remained after operation but did not require analgesics and he was able to resume an active and useful life (Case 3 in the appendix).

A thalamic syndrome of intolerable pain in the left side of the face occurred after a thrombosis of the left posterior inferior cerebellar artery in an elderly patient with severe hypertension. This pain, caused by damage to the left quintothalamic tract, had persisted for four years and failed to respond to large doses of analgesics and sedatives. Destruction of the right arcuate nucleus of the thalamus completely abolished the pain (Case 4 in the appendix).

In three patients the pain was considered to be emotionally elaborated and the lesion was placed in the dorso-medial nuclei of the thalamus on both sides. A young woman had suffered from intractable pain in the scar of a radical mastectomy for three years. She attempted suicide after a dorsal rhizotomy failed to relieve this pain. It was completely abolished by coagulation of both dorso-medial nuclei and no personality disorder nor intellectual deficit resulted from this operation (Case 5 in the appendix).



FIG. X. Coronal macroscopic section of brain of Case 1, showing electrolytic lesion in posterior ventral nucleous of thalamus.

An elderly man with a twenty-year history of a mild quadriparesis due to cervical spondylosis had complained of vague pains in every part of his body and of unbearable spasms in his limbs for many years. After destruction of both dorso-medial nuclei he continued to complain of vague aches and pains, but seemed quite unconcerned about them and never asked for analgesics. He learned to walk with the aid of crutches, but fell a few days after discharge from hospital and fractured his femur (Case 6 in the appendix).

Our last patient had suffered from an atypical neuralgia of the right side of the

face for fifteen years. Dental extractions, operations to the antrum and nose, ligation of the external carotid artery, alcohol injection into the infra-orbital nerve, and even intracranial section of the maxillary and mandibular nerves had failed to influence this pain. Electro-convulsive therapy relieved it for four weeks only. After coagulation of both dorsomedial nuclei he continues to complain of this pain, but is indifferent about it and no longer requires analgesics. No evident intellectual impairment resulted from operation and he has returned to work (Case 7 in the appendix).

Further clinical details of these patients are described in an appendix to this paper.

Our small number of cases and the results of overseas surgeons cited from the literature, do not yet permit a statistical assessment of the value of these operations. The success achieved in individual patients proves, however, that stereotactic brain surgery can relieve pain, which by its nature and pathogenesis has in the past been intractable.

#### ACKNOWLEDGEMENTS

We are indebted to Professor T. Riechert for his valuable advice by correspondence and for permission to publish this description and photographs of his instrument.

Drs. E. A. Spiegel and H. T. Wycis have kindly granted us permission to reproduce a plate from their book on stereo-encephalotomy.

Our thanks are due to Dr. J. Kalokerinos and to the senior resident medical officers of the X-ray department of the Royal North Shore Hospital of Sydney for their generous help in providing the important radiological facilities for these operations.

Dr. C. Graham performed the autopsy on Case 1, and Dr. Brian Turner prepared the sections of the brain.

#### APPENDIX

Summaries of case histories of the patients reviewed in this paper.

#### Case 1

W.J.H., a male, aet. 71, had an attack of left ophthalmic herpes zoster in February, 1958. During the following six months he suffered from constant and severe pain in the left forehead and eye, which failed to respond to analgesics and demanded permanent hospital care.

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nus the Examination showed the typical post-herpetic scarring of the forehead and analgesia over the ophthalmic division of left trigeminal nerve.

X-ray of the chest revealed chronic inflammatory changes in both upper lobes.

On 25th Oct., 1958, the right arcuate nucleus of the thalamus was destroyed by electro-coagulation. During operation analgesia of the entire left half of the face, and hypoalgesia of the left limbs and trunk developed. After operation the patient was mentally confused for two days, but then improved and stated that the pain in his left forehead had ceased, but that he still had some discomfort in his left eye.

Four days after operation he had an extensive anterior cardiac infarction, confirmed by E.C.G.,

and he died on the following day,

Autopsy demonstrated the lesion in the right arcuate nucleus. It was larger than intended due to surrounding haemorrhage and involved some of the lateral part of the posterior ventral nucleus of the thalamus, but did not encroach on the internal capsule. Occlusion of the left coronary artery was confirmed and fibro-caseous tuberculosis was found in both lungs.

#### Case 2

L.H., a woman of 69 years, suffered from left ophthalmic herpes zoster with severe involvement of the cornea at the end of March, 1959. She was left with an intractable post-herpetic neuralgia, which had failed to respond to deep X-ray therapy to the left Gasserian ganglion, and to very large doses of chlorpromazine and analgesics. The left eye was inflamed and closed by swollen lids and she would not permit anyone to touch her left forehead or eye-lids.

At a stereotactic operation on 30th June, 1959, the right arcuate nucleus of the thalamus was destroyed. Analgesia of the left side of the face and hypoalgesia of the left half of the body and limbs were observed during operation. Numbness of the left half of the tongue, mouth, lips and of the left limbs subsided three weeks after operation. Hypoalgesia over the ophthalmic division of the left trigeminal nerve remained. Pain in the left forehead was completely abolished, but some discomfort in the left eye persisted.

When examined seven weeks after operation, she had resumed an active life, but still required an average of four aspirin and codein tablets per day for the relief of pain in her left eye. The left pupil was large, irregular and failed to react to light. She had complete analgesia to all modalities of sensation over the ophthalmic division of the left trigeminal nerve, and a moderate hypoalgesia in the remaining two divisions. The left corneal reflex, however, was only slightly diminished. Cutaneous, proprioceptive and discriminative sensation were normal in the left limbs and trunk. Muscle tone, power and co-ordination remained normal in the left half of the face and left limbs.

It is thought that some of the residual pain in the eye may be mediated by afferent sympathetic fibres, which do not terminate in the arcuate nucleus,

#### Case 3

C.F., a man aged 69 years, had suffered from herpes zoster involving the fourth and fifth thoracic dermatomes on the left side in 1957. Constant and severe post-herpetic neuralgia persisted for two years, interfered with sleep, prevented him from working, and was not relieved by large doses of analgesics and sedatives.

Scars remained over the affected dermatomes and he had hyperaesthesia over the left half of the back of his chest, extending from the neck to the lower thoracic region, and would not permit this area to be touched.

The lateral part of the posterior ventral nucleus of the right thalamus was destroyed by diathermy coagulation on 16th May, 1959. During the operation he retained the appreciation of pin-prick and touch on the left side of his body and limbs, but he always neglected the left side when both sides were stimulated simultaneously.

After operation he was mentally confused for ten days, and developed a mild left hemiparesis, which recovered completely after one week. He continued to complain of slight pain in the affected dermatomes, but his hyperaesthesia was abolished and he had no discomfort when the affected area was touched or rubbed.

Seven weeks after operation he reported only slight discomfort in the post-herpetic scar, which no longer required any analgesics or sedatives. He had resumed an active life in his home and garden. Cutaneous and proprioceptive sensation remained intact in the left half of the trunk and limbs, but some neglect of the left side on bilateral simultaneous stimulation persisted,

#### Case 4

C.M., a man aged 72 years, with long standing hypertension, had a thrombosis of the left posterior inferior cerebellar artery in August, 1955. This caused vertigo, ataxia, dysarthria, and numbness of the left side of his face and of the right half of his trunk and limbs. Ataxia and dysarthria improved within six months, Early in 1956 the numbness of his face subsided and was replaced by a constant burning pain and hyperaesthesia over the left half of his forehead and face. He could not even tolerate the contact of a soft pillow with the affected side. This spontaneous burning pain of thalamic origin persisted for three and a half years and made his life a misery. Large doses of chlorpromazine, up to 400 mgm. daily, failed to relieve it, and he was in the habit of taking an average of twelve A.P.C. powders per day and developed methaemoglobinaemia.

Examination showed a left Horner's syndrome, depression of the left corneal reflex, moderate hypologiesia over all three divisions of the left trigeminal nerve, and a mild hypologiesia of the right limbs and right half of the trunk. Only slight ataxia of the left limbs remained. His heart was enlarged, and the blood pressure elevated to 220 mm. of mercury systolic and 120 mm. of mercury diastolic.

On 11th June, 1959, the arcuate nucleus of the right thalamus was destroyed by diathermy coagulation. Because of methaemoglobinaemia the entire operation was performed under local anaesthesia. Complete analgesia of the left half of the face and hypoalgesia of the left limbs developed during

the operation. No post-operative complications occurred and the pain in his face was completely abolished.

On examination nine weeks after operation he reported complete freedom from pain and no longer required any analgesics. Normal cutaneous, proprioceptive and discriminative sensation had returned to his left limbs and he had no inattention on bilateral simultaneous stimulation, The left corneal reflex remained depressed, He had a hypoalgesia to all modalities of sensation over the left half of the forehead and face, of the same degree and extent as before operation. No change in his mental faculties, or in the power and co-ordination of his left limbs had resulted from the operation.

#### Case 5

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E.F., a housewife aged 43 years, had a left radical mastectomy in 1955. Histological examination showed a hormonal mastopathy and excluded carcinoma. The wound failed to heal and was repaired with skin grafts. Ever since this operation she had suffered from a persistent pain in the scar, unrelieved by analgesic drugs and sedatives. She was unable to care for her home and children and required almost constant hospital attention. Rhizotomy of the second to fifth thoracic posterior roots relieved her pain for three weeks only. She was depressed, had attempted suicide on one occasion and was given electro-convulsive therapy without improvement.

Both right and left dorso-medial nuclei of the thalamus were destroyed by diathermy coagulation on 11th Oct., 1958. She was confused, disorientated and incontinent for ten days after operation. By the fourteenth day she was mentally alert and free from pain.

When reviewed five months after operation, she reported complete freedom from pain and had resumed her normal domestic duties. Her relatives had not observed any personality deterioration or intellectual impairment and both she and her family were delighted with the result.

#### Case 6

W.M., a man of 66 years, had suffered from a mild spastic quadriparesis, without sensory impairment or sphincter involvement, for twenty years. Extensive investigations showed that this was due to cervical spondylosis, but many years of orthopaedic and palliative treatment had failed to help him. He complained of constant and intolerable pain in his trunk and limbs and was subject to painful muscle spasms of his arms and legs. He became depressed and thought of suicide.

On 26th July, 1958, the dorso-medial nuclei of the thalamus on both sides were destroyed, A large lesion was made first in the right dorso-medial nucleus and he remained mentally alert and well orientated. After twenty seconds of diathermy coagulation of the left nucleus he became mentally confused.

He was irrational and incontinent for four weeks after operation, but then regained sphincter control and learned to walk on crutches. When questioned, he still complained of various aches and pains, but seemed quite unconcerned about them and never asked for analgesics. His muscle spasms ceased,

Seventeen days after discharge from hospital he fell and fractured the neck of his right femur. He was readmitted and remained in hospital for a

further five months. During this time he was occasionally depressed, but complained only once of muscle spasms. He did not demand analgesics, participated in occupational therapy and gradually learned to walk again with the aid of crutches.

#### Case 7

V.E.W., a male, aet. 40, had suffered from a constant pain in his right cheek and behind the right eye for fifteen years. Any excitement or emotional stress aggravated this pain. He had consulted many doctors, but repeated antral lavages, submucous resection of the nasal septum, extraction of all his teeth, ligation of the right external carotid artery, and alcohol injection of the right infra-orbital nerve failed to give him any relief. In June, 1957, the second and third divisions of the right trigeminal nerve were cut by a Frazier operation, resulting in objective sensory loss over the appropriate area. He was free from pain for only three weeks after this operation, and then began to complain of a constant ache in the right lower gum and jaw, A few months later this spread to the upper gum and he again demanded large doses of analgesic tablets and frequent pethidine injections. Chlorpromazine, in a dose of 300 mgm. daily, did not help him.

Psychiatrists thought him depressed, and in July, 1958, he had a course of electro-convulsive therapy, which abolished his pain for four weeks. When it recurred, further E.C.T. was not effective. In April, 1959, he claimed that the pain had increased in severity to a degree where he could no longer continue his work as a clerk.

On 2nd July, 1959, both right and left dorsomedial nuclei of the thalamus were destroyed by diathermy coagulation. He was confused for one day only, and was discharged from hospital one week after operation.

Three weeks later pain in the right side of his face recurred, but he described it with remarkable indifference and did not ask for analgesics, No intellectual impairment was evident after operation and he returned to his clerical work.

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# NECROTIZING ENTERITIS\*

By W. W. WOODWARD

Launceston

PRESENTING as apparently spontaneous shock or as diarrhoea or as both this catastrophe is nearly always fatal. At necropsy there is diffuse or patchy necrosis in almost any region of the alimentary tube but usually in jejunum, ileum or colon. mucosa is the layer affected but at times, as in the third patient below, all layers are dead. The necrotic areas are yellow or brown, irregular in outline and sharply demarcated. The bowel wall may be congested or even oedematous. The lumen may show bloodstained fluid and/or casts of sloughed mucosa (Dawson-Edwards and Morrissey, 1955). Casts account for an alternative title, that of pseudomembranous enterocolitis.

As in the first necropsy below the serosa may show no change, or be oedematous, or congested as in the second necropsy below, or covered with a fibrinous exudate, or involved in patches of gangrene as in the third patient.

### CASE REPORTS

#### Case 1

(Launceston General Hospital — No. 47908.) G.P.C., a male aged 47, was admitted to the care of Dr. M. W. Fletcher in September, 1951, because of dyspnoea and atrial fibrillation. Two years earlier it had been necessary to stop using the leaf of digitalis because in ordinary dosage it did not control the fibrillation while larger doses gave vomiting and diarrhoea. Digitoxin was used instead.

For years he had suffered bouts of upper abdominal pain and two days after admission had another such attack with vomiting, but this time there escaped from his rectum pink fluid, microscopy of which showed numerous red blood cells with relatively few polymorphs. The digitoxin now in turn was ceased.

During his last twenty-four hours he passed no urine and we then noted fixed hypogastric dullness to percussion. Catheterization however yielded only 2.0 ml. of clear urine. Accordingly I entered into the progress notes that the dullness was unexplained,

adding that shock was severe, the abdomen extremely tender, bowel sounds absent and my diagnosis was then mesenteric vascular occlusion. Less than an hour later, seventeen hours from the rectal loss of blood-stained fluid, the patient died.

A necropsy was done by Dr. M. P. K. Shoobridge in the company of Dr. Fletcher and myself. In the peritoneal sac were 20.0 ml. of clear straw-coloured fluid. Throughout the small bowel the mucosa showed many separate congested areas and there were 280 ml. of thick blood-stained fluid in the distal third of the ileum; hence presumably the hypogastric dullness. In the terminal 40 cm. of ileum the mucosa was rough and coated with slough. Despite this the overlying serosa was normal. The superior mesenteric artery and vein were patent.

# Case 2

(Launceston General Hospital — Unit Record 2199.) Miss E.J., though aged 76, was in relatively good condition when in June, 1956, she underwent pancreato-duodenectomy. After operation penicillin and streptomycin were ordered. Her condition remained satisfactory until 8 p.m. on the second postoperative night when her blood pressure fell steeply: within an hour she was moribund and at 3.15 a.m. she died.

Necropsy by Dr. P. Hamilton in my company revealed a 35 cm. length of purple lower jejunum and two similar lengths of purple ileum. On opening these congested portions we saw in the mucosa many scattered brown patches of necrosis. All the major branches of the superior mesenteric artery and vein were patent.

#### Case 3

(Launceston General Hospital — Unit Record 10672.) H.L.R., a male aged 56, was admitted at night in August, 1958. He had felt well until noon that day when he had a sudden urge to open his bowel. He strained without success, took a dose of castor oil and developed slowly intensifying midline upper abdominal pain. As he lay in bed his knees were drawn up, his respirations short and grunting.

At 12.30 a.m. laparotomy by Mr. R. P. Booth revealed intra-peritoneal brown fluid and patches of gangrene in the wall of the large bowel from ascending colon to sigmoid. The patient's condition was desperate: the colon was exteriorized and a caecostomy made. Intravenous hydrocortisone sodium

<sup>\*</sup>Received for publication, 30th November, 1959.

succinate 133.7 mg. (i.e. "Solu-Cortef' 100 mg.) was used but, despite this and noradrenaline, shock was irreversible. Twenty-nine and one-half hours from the onset of his pain the patient died.

Necropsy by Dr. B. Hartley revealed: (1) brown fluid in the peritoneal sac, (2) patches of gangrene varying in length from 1.25 to 7.5 cm. along the colon and small bowel, and (3) patent inferior and superior mesenteric vessels.

The main features are now tabulated.

## (2) Treatment

Non-operative: The usual measures to combat rapidly progressive shock are mentioned by Kay, Richards and Watson (1958) but all their 16 patients died. Unless a recognizable cast were to be seen (Muir, 1957) a survivor would not provide evidence of having had the disease and it is possible, therefore, that energetic medical treatment has at times succeeded.

	Previous condition	Antibiotics received	Clinical features	Time from assumed onset to death	Site of bowel affected	Appearance of bowel from serosal aspect	
First patient	Cardiac failure, atrial fibrilla- tion, intolerance of digitalis	Nil	Passage of blood- stained fluid per rectum. Lower abdomen dull to percussion despite an empty bladder and absence of ascites	17 hours	Ileum	Normal	
Second patient	Obstructive jaundice, pancreato- duodenectomy	Post-operative penicillin and strepto- mycin	Sudden arterial hypotension 30 hours after operation	7½ hours	Jejunum and ileum	Three long regions of purple congested small bowel	
Third patient	Nil	Nil	Epigastric colic	29½ hours	At necropsy: colon and small bowel	At laparotomy: patches of colonic gangrene	

#### COMMENT

#### (1) Cause unknown

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Necrosis of bowel mucosa with membranous sloughing is not new. Finney (1893) ascribed a death after gastro-enterostomy to "diphtheritic" colitis and "croupous" enteritis was recognized by Muir (1941), i.e. before antibiotics were in therapeutic use. Necrotic enteritis follows bowel surgery (Williams and Pullan, 1953), surgery outside the abdomen (Dixon and Weismann, 1948), or occurs without preceding surgery (Penner and Bernheim, 1939). Zeissler and Rassfeld-Sternberg (1949) accused a previously unrecognized anaerobe of the Clostridium welchii group. Cook, Elliott, Elliott-Smith, Frisby and Gardner (1957) propose that staphylococci would be found more if sought more, a proposition considered by Penman and Pullan (1958). Further discussion is given by Bruce (1955), Hayward (1959) and another Australian example of the disease is described by Webb (1959). No one theory accounts for all cases.

Operative: Of 18 patients with necrotic jejunitis reported by Fick and Wolken (1949) 16 died: the two survivors were treated by resection of the affected portion of jejunum. Hence operation offers the only hope known.

#### SUMMARY

- Three fatal examples of necrotic enteritis are reported. In two patients the disease followed neither surgery nor antibiotic therapy.
- The lower abdomen of one patient was dull to percussion though the urinary bladder was empty and ascites absent at necropsy. For want of other explanation this is ascribed to fluid in the lumen and congestion in the wall of the ileum.
- At necropsy on that patient the disease was not visible from the serosal aspect of the bowel. Hence at laparatomy recognition might require palpation of the bowel.

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# RANULA\*

# By MILROY PAUL

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It is felt that the subject of ranula has not received the attention that it merits. Our experience and conclusions from treatment in our series may be of interest to others. Thirteen cases of ranula in adults and 5 cases of ranula in young children have been treated over a period of twenty-four years. In 16 cases the ranula was completely excised, therefore disproving the general belief that the dissection of a ranula often proves impracticable because of its thin transparent fragile walls. Marsupialization into the floor of the mouth was done in 2 cases but, looking back on these cases in retrospect, it is felt that they also could have been dissected out.

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The complete excision of a ranula proved to be practicable because of the cyst wall being loosely attached in its bed, to the cyst having no attachment to Wharton's duct or to the sublingual gland, and to the thin fragile transparent wall manifesting a remarkable tolerance to the blunt manipulations needed to enucleate it out of its bed. The cyst wall may be ruptured by coming into contact with the edges of the teeth while being delivered out of the mouth or to the cyst being wounded by instruments during dissection. Avoidance of these hazards makes the complete excision of every ranula a feasible operation.

#### WHAT IS A RANULA?

The name ranula (rana Latin; a frog) gives no clue to the origin of the condition. The significance of the name has been forgotten, and whether the name refers to a ranula looking like the stomach of a frog (Charles Bell) or to some other fanciful resemblance to a frog is now a matter of conjecture. There is however general agreement as to the features of a ranula. A ranula may be defined as a transparent cyst in the floor of the mouth. The property of transparency excludes the dermoids from the category of the ranulas.

A ranula occupies a constant position in the floor of the mouth. It is an ovoid cyst occupying the lateral sulcus between the tongue and the mandible. Its anterior pole reaches up to and even a little beyond the midline while its posterior pole extends for a variable distance backwards. The constant position of the ranulas suggests that they originate from some structure in the sulcus between the tongue and the mandible.

Many writers (Hamilton Bailey, 1959; Fitzwilliams, 1927; Curtis, 1898; Eve, 1889) have described transparent cysts on the under surface of the tongue as ranulas. These cysts of the tongue are as constant in position as are the ranulas. They are ovoid cysts lateral to the middle line with long axes parallel to the long axis of the tongue. The site of these cysts suggests that they originate in the glands of Blandin and Nuhn.

Although a cyst on the under surface of the tongue is in the floor of the mouth, these cysts are clearly different from the ranulas. They never extend into the sulcus between the tongue and the mandible, while the ranulas on the other hand never extend upwards on to the under surface of the tongue. It would appear improbable that ranulas originate, as was supposed by Von Recklinghausen (1881), in the glands of Blandin and Nuhn. The cysts on the under surface of the tongue are transparent cysts, but so are the mucous cysts of the lips and cheeks. They are all cysts of the same type, but they are not ranulas on that account.

#### Contents

What structure in the sulcus between the mandible and the tongue could be the origin of the ranulas? The sublingual salivary gland and the duct of the submaxillary salivary gland are both submucous in this sulcus. Could a ranula be a retention cyst of either of these salivary glands? Gmelin found that the fluid in a ranula had no "sulpho-cyanic acid alkali" and that it was rich in albumin

<sup>\*</sup>Received for publication 13th January, 1960.

which is not a constituent of saliva. Lockwood (1882) found that ranula fluid did not convert starch to glucose, it was soluble in distilled water and was precipitated by acetic acid. It did not give a claret colour with ferric perchloride. Eve also found no potassium sulpho-cyanide in the fluid of ranulas. A ranula could not therefore be a retention cyst of a salivary gland. Thompson (1920) showed by dissection that both Wharton's duct and the sublingual gland were separate from and superficial to the ranula.

The ranula fluid is not unlike the fluid in a semimembranosus bursa and the fluid in the ganglia of tendon sheaths. Could the ranulas therefore originate from a bursa sited in the sulcus between the tongue and the mandible? Stromeyer was so convinced that there must be a bursa at this site, that he persuaded Fleishmann of Erlagen to investigate this point. Fleishmann (1841) wrote "If from one or other side of the fraenum, one separates the mucous membrane from the tongue, one finds close to the fraenum, resting on the genioglossus muscle, behind the duct of Wharton and the ducts of Rivini, a small mucous bursa, oval shaped, divided into cavities by cellular partitions, the sublingual bursa, the existence of which it is important to be acquainted with for a knowledge of ranula". Fleishmann's bursa could not be found by other observers (Thompson, 1920) and its existence as an anatomical structure is now discounted.

Thompson found histological studies of the walls of a ranula to be unsatisfactory. He was unable to find epithelium in many of the cases and when found it was of the tesselated variety. For the most part the wall was made up of dense fibrous tissue with large lymphatic channels in it. He concluded that the structure of the wall conformed to that of the branchiogenetic cysts and postulated that ranulas are branchial cysts carried forward by the migration of the hypoglossal and branchial arch muscles. The fluid of a branchial cyst however bears a striking resemblance to tuberculous pus (Hamilton Bailey, 1959) and it is quite unlike the mucoid jelly filling a ranula. Moreover a branchial cyst is nearly always lined by squamous epithelium. If a ranula were derived from a branchial origin, its posterior pole would be its most constant point and it would extend for a variable distance forwards.

The posterior pole of a ranula on the other hand is its most variable point while its anterior pole is very nearly constant.

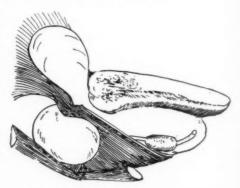


FIG. I. Ranula with an extension into the submaxillary triangle of the neck and an extension lateral to the superior pharyngeal muscle.

Where then could a ranula originate, and what is its nature? The fluid in a ranula is similar to the fluid in the transparent mucous cysts of the lips and cheeks. These cysts of the lips and cheeks are believed to be retention cysts of mucous glands. The cysts in the cheeks are larger than the cysts in the lips and this may be because they are able to enlarge without resistance. A retention cyst in a mucous gland in the sulcus between the tongue and the mandible would also be free to enlarge along this sulcus, and if it did, it would assume an ovoid shape, it would reach up to and slightly beyond the midline, but would be prevented by the fraenum from extending much more beyond the middle line. Posteriorly the cyst could extend up to the end of the sulcus between the tongue and the mandible. It could then burrow upwards between the mandible and the pharyngeal wall or downwards behind the posterior border of the mylohyoid muscle into the submaxillary triangle in the neck, or it could do both of these things (Fig. I). Ranulas have been found with these extensions, but every ranula of this kind also occupied the sulcus between the tongue and the mandible. The evidence suggests that a ranula enlarges in this way because of the looseness of the tissues at these sites. The mucoid jelly content of the ranula is the same as the mucoid jelly content of the retention cysts of mucous glands, of the semimembranous bursa, and of the ganglia of tendon sheaths. The ranula cannot originate in a bursa. It must therefore be a retention cyst of a mucous gland attaining its large size because of the loose cellular tissues in the sulcus between the tongue and the mandible.

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# TREATMENT

A ranula constitutes a deformity and a disability and will increase in size. The ideal treatment for a ranula would be excision of the whole cyst with its walls unruptured. Unfortunately the thin transparent walls are so fragile that most surgeons have found that excision of a ranula with its walls intact is impracticable and in several recorded cases the cyst wall has only been incised or partially excised. Any operation which leaves behind portions of the cyst wall would be followed by a recurrence of the cyst. Handfield-Jones and Porritt (1957) advise that, as excision of the cyst is not possible, the projecting wall of the cyst be excised flush with the floor of the mouth and the remaining portion of the cyst wall be destroyed by diathermy. Hamilton Bailey (1959) states that as the cyst would usually rupture before it could be excised, it should be deflated by aspiration of some of its fluid content and then dissected out of its bed. If aspiration was not possible on account of the fluid being too viscid, the projecting part of the cyst wall could be excised and the lower part of the cyst wall could then be marsupialized by suturing its edges to the mucous membrane of the floor of the mouth. These approaches to the problem of treatment will make it evident that ranulas are likely to recur after surgical operations. Can the problem be solved?

The wall of a ranula is thin and loosely attached to the tissues (Thompson 1920). Why does the wall rupture when the cyst is being excised? The buccal mucosa lies immediately over the cyst wall and incisions on this mucosa are likely to open the cyst wall. Even if this danger be avoided, the cyst could still rupture while it is being dislocated out of its bed. A careful study of this part of the operation demonstrated that the rupture occurred on drawing the cyst wall over the teeth of the mandible. The teeth wounded the cyst wall. If this danger be avoided the unruptured ranula can be manipulated without undue risk of injury and it is possible to dissect out a ranula with its walls intact.

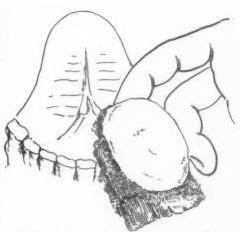


FIG. II. Enucleation of a ranula from its bed by blunt finger dissection. Note that the cyst is protected from injury by a square of gauze placed over the teeth.

A new operation for the excision of a ranula

Wharton's duct runs over the surface of the ranula with the sublingual gland lateral to it. The ranula projects into the floor of the mouth, and it is difficult to divide the mucous membrane overlying a ranula without wounding its wall. An incision can however be made through the buccal mucous membrane in the linear groove between the side of the tongue and the ranula. A finger can be inserted through this incision between the ranula and the hyoglossus muscle and the cyst wall separated from its bed. The finger can now free the cyst wall at its anterior pole. The buccal mucous membrane can next be dissected off the wall of the ranula with greater ease than if the dissection were made before the ranula was mobilized in its bed. Wharton's duct and the sublingual gland remain attached to the flap of buccal mucous membrane during the dissection and they will not be injured. The final step in the operation is freeing the under surface of the ranula from the mylohyoid muscle. To do this the finger is hooked under the ranula and the ranula is lifted further and further out of the mouth. At this stage the teeth on the mandible should be covered with gauze, and care must be taken not to push the cyst over bare teeth (Fig. II). With care and patience, the cyst can be enucleated entire out of its bed. There is no bleeding. One or two interrupted sutures suffice to appose the edges of the incision in the buccal mucous membrane.

#### SUMMARY OF CASES

Initials	Age	Sex	Date of Admission	Duration of ranula	Remarks		
P.	21	M	14/12/36	1 month	Dissected out entire, cyst burst.		
C.H.F.	18	F	24/10/38	1 month	Dissected out piecemeal.		
H.E.	22	M	26/4/44	3 days	Anterior wall excised.		
B.S.	9	M	20/11/44		Dissected out entire.		
W.R.	35	M	28/10/47	6 months	Dissected out entire, cyst burst.		
N.N.	12	M	28/9/48	2 years	Dissected out entire.		
M,	16	M	25/4/50		Dissected out entire, cyst extended int submaxillary triangle, removed through incision in neck.		
D.	48	M	1/5/51	2 months	Excised completely.		
N.C.P.	17	M	5/8/52	2 weeks	Dissected out entire.		
G.R.B.	30	M	26/3/58	12 months	Dissected out entire.		
G.B.D.	19	M	26/5/59	2 months	Dissected out entire.		
R.M.P.	28	M	4/8/59	1 month	Dissected out entire.		
P.S.	12	F		8 months	Dissected out entire.		
I.	7	F	12/11/46	6 months	Dissected out entire, cyst burst.		
A.	6	F	12/11/46		Dissected out entire.		
B.	8	F	14/9/58	2 weeks	Dissected out entire, cyst burst.		
S.	8	F	5/6/57	8 months	Cyst marsupialized.		
M.O.	211/12	F			Dissected out entire, cyst burst.		

The patients above the line were treated at the General Hospital, Colombo; those below the line at the Lady Ridgeway Hospital for Children, Colombo.

# ACKNOWLEDGEMENTS

The figures illustrating this article were drawn by Mr. A. S. Mahawatte, Photographic Artist, University of Ceylon. Mr. A. B. Fernando and Dr. M. Ramachandran very kindly collected the case records of ranula at the General Hospital, Colombo and at the Lady Ridgeway Hospital for Children, Colombo, respectively.

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# PRIMARY DUODENAL DIVERTICULA AND THEIR PROBLEMS\*

By DANIEL LANE

Brishane

"The golden rule is that there are no golden rules" - Shaw.

THE more I see and study primary duodenal diverticula, the more uncertain I feel of their behaviour. From my study of hospital statistics and private records it is obvious that few of us have any great operative experience of true primary diverticula.

In general, all orthodox surgeons eschew operation in duodenal diverticulum because it would be meddlesome, dangerous and quite unlikely to relieve symptoms. But there must be some diverticula which require operation. It is my purpose in writing this paper to present my experience of several diverticula which have required operation within the past three years. I have also reviewed the recent statistics of the Brisbane General and Mater Hospitals.

#### Statistics

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From Table 1 it is seen that 28 cases were admitted to hospital for treatment of symptoms considered due to the diverticula. During the past ten years it has been possible to collect only 8 cases in Brisbane where excision of diverticula has been performed. There have been a few other cases but adequate clinical records are not available. Thus it is apparent that the operation for removal of a duodenal diverticulum is extremely rare in Brisbane (population approximately 500,000).

The average age group, 60 years, is consistent with other reports. It was interesting to note that a high proportion of New Australians was included. Many of them are unable to speak English well and it is necessary to take extra care when attempting to analyze their symptoms.

It is easy to ascribe the symptoms to a duodenal diverticulum accidentally noted during routine radiological studies. Sometimes a more serious disease such as carcinoma can be overlooked (Case 3).

Of the 28 cases admitted to hospital, 9 had undergone prior cholecystectomy (about 31 per cent.) which further emphasizes that there are many unjustifiable cholecystectomies still being performed in Australia. In many of the gall-bladders removed, calculi were absent and pathological change was minimal.

We must be very chary of removing an apparently normal gall-bladder in an attempt to cure vague upper abdominal symptoms. It would seem unnecessary to state that all patients with equivocal cholecystogram reports should have a barium meal as a routine in an attempt to exclude such conditions as duodenal diverticulum which may produce symptoms strongly suggestive of gall-bladder disease.

The majority of the patients studied suffered from symptoms which could easily be interpreted as biliary in origin.

There was no appreciable sex difference in this series.

Six cases presented with either haematemesis or melaena, the majority with the latter. This was surprisingly high and it remains doubtful whether the diverticula were the cause of the haemorrhage in many instances.

It was noted that in many cases repeated barium meal examinations were required in order to show the diverticula. Thus it would seem necessary to repeat the barium meal at least once in any case with vague dyspeptic symptoms and in whom all other investigations have proved negative. Sometimes it is wise to change the radiologist.

The majority of the cases reviewed were treated by diet, rest and an ulcer regime which relieved symptoms.

<sup>\*</sup>Received for publication, 24th August, 1959.

It has been impossible to ascertain exactly the proportion of cases cured of symptoms by operation but I suspect that it was less than 50 per cent. Duodenal diverticulum is in a similar category to hiatus hernia and requires conservative treatment for as long as possible.

I now wish to illustrate from case records certain aspects of primary duodenal diverticula which warrant close attention. Laparotomy performed on 10th August, 1956, revealed a thin-walled diverticulum about 2 inches in diameter arising near the ampullary region of the duodenum. The neck of the diverticulum was very narrow and it appeared that emptying of the diverticulum was difficult. The common bile duct was thick-walled and dilated, but a probe could be passed readily into the duodenum. It seemed reasonable that the diverticulum was the cause of the recurrent episodes of jaundice by virtue of obstruction to the common bile duct. The diverticulum was removed with particular care owing to its proximity to the

#### TABLE 1

# ANALYSIS OF 28 CASES OF DUODENAL DIVERTICULA REQUIRING ADMISSION TO HOSPITAL FOR TREATMENT OF SYMPTOMS DIRECTLY RELATED TO THE DIVERTICULA

	Total number of cases reviewed	Cases of direct operations on Diverticula	Operative Mortality	Average Age	Sex Ratio	Prior Cholecyst- ectomy	Dominant Symptoms	Haematemesis or Melaena
28 <	18 Brisbane Hospital 1947–1957 5 Mater Hospital 1947–1957 4 Private (Personal) 1956–1959 1 Private (Borrowed) 1957.	8	1	60 years	Female Male	9	Vague epigas- tric discomfort and flatulence, sometimes re- lated to meals	6

#### Case 1

# Diverticulum causing common bile duct obstruction

Mr. F.P., aged 61 years, presented privately on 23rd July, 1956, with a history of jaundice and fever during the preceding week. He had also experienced colicky upper abdominal pain and vomiting. His past history was interesting. In 1947 he underwent cholecystectomy for acute cholecystitis; there were gangrenous patches in the gall-bladder wall and several calculi were present. He had a stormy convalescence and developed a biliary fistula. Six weeks after his first operation he was re-explored but no obstruction of the common bile duct could be found. The fistula ultimately closed and he remained well until 1949 when he suffered a further attack of colic. He next presented to his local doctor in February, 1956, with a melaena which was thought due to diverticulitis of the distal colon; the barium meal was negative. Since then he had experienced at least one attack of jaundice lasting several days.

Examination revealed a sick, deeply jaundiced man with the swinging temperature of a typical cholangitis. Apart from an incisional hernia, the chief abdominal finding was an enlarged hard liver. There were no other relevant findings.

Serum bilirubin on 24th July, 1956, was 8.5 mgms. per 100 cc, which gradually fell to 0.7 mgm. per 100 cc. on 30th July 1956. Routine liver function tests were consistent with an obstructive jaundice. ampulla. A T-tube was left in the common bile duct. The post-operative cholangiogram showed some narrowing of the distal end of the common bile duct but the dye flowed freely into the duodenum. The diverticulum showed evidence of inflammation. The patient made an uneventful recovery.

#### Comment

This patient has remained well but there is always the possibility of further trouble arising as a result of cirrhosis subsequent upon the several episodes of obstructive jaundice experienced.

It would seem reasonable to consider that the melaena was also due to the duodenal diverticulum. The discovery of the diverticulum was iortuitous and was found as a result of preliminary mobilization of the duodenum. This case teaches us the lesson that duodenal diverticulum must be considered as a cause of obscure obstructive jaundice.

#### Case 2

### Diverticulum and the danger of operation

Mrs. A.B., aged 58 years, was seen privately on 23rd April, 1958, with a history of flatulent dyspepsia for many years. She claimed that her cructations at meal time were becoming a source of embarrassment.

Some of her symptoms were relieved by taking soda water. She had also been treated for a hypochromic anaemia which remained unaltered. Appendicectomy for acute appendicitis had been performed six months previously.

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FIG. I. The barium meal appearance of Case 2 showing the two diverticula.

Examination of the patient revealed a wellnourished, pale woman who was slightly hypertensive. Barium enema showed sigmoid diverticula. Barium meal revealed two large diverticula arising in the ampullary region of the duodenum. There was considerable stasis of barium (Figs. I and II). Cholecystogram was considered normal.

The patient ultimately decided that she wanted the diverticula removed. Laparotomy was performed on 13th May, 1958. Two large diverticula were found arising from the medial wall of the second part of the duodenum. The orifice of the proximal diverticulum was very close to the ampulla (Fig. III). It was necessary to open the common bile duct and insert a probe during excision of the proximal diverticulum. The diverticula were excised and the duodenal wall closed in two layers. A T-tube was inserted into the common bile duct and a drain placed behind the duodenum.

The patient seemed well until 17th May, 1958, when her condition appeared to deteriorate. A diagnosis of pancreatitis was made and supported by a moderate rise in the urinary diastase (150 Wohlgemuth units).

On 19th May, 1958, she appeared much improved. Her behaviour was rational and she had passed flatus. It was decided to remove the Ryle's tube but to continue with intravenous therapy for another day. On the following day after taking some jelly and junket she died suddenly.

Autopsy revealed pancreatitis localized to the right paracolic gutter but without evidence of general peritonitis. The duodenal closure was quite secure. The heart was dilated and there was a bilateral pleural effusion. It was considered by the pathologist that death was due to circulatory failure aggravated by intravenous therapy. The pancreatitis was an important contributory cause of death.

In retrospect we found that on one day the patient had received much more intravenous fluid than ordered



FIG. II. The barium meal of Case 2 showing stasis of barium within the diverticula.

#### Comment

The main lesson to be learnt from this patient is the grave risk of pancreatitis in operation on duodenal diverticula. Both these diverticula were in close contact with the pancreas and required dissection of pancreatic tissue. This is a hazardous procedure because even minimal handling of the pancreas can initiate severe pancreatitis.

The patient insisted upon operation but it would have been better to have continued with conservative treatment until more severe symptoms developed. It is in such cases that it seems better to err on the side of conservatism.

In this patient the retro-duodenal corrugated rubber drain proved quite valueless and gave no indication of the intra-peritoneal complication. Intravenous therapy is still a dangerous form of therapy if not carefully supervised by experts. This patient may have

survived but for the excess intravenous fluids administered at a critical stage.

Case 3

Diverticulum masking symptoms of serious disease

Mrs. E.W., aged 65, was seen privately on 12th April, 1958, complaining of discomfort in the left groin. There was also a history of indigestion after meals and some pain in the right hypochondrium. She had suffered from a stroke some years previously but had made a good recovery.



FIG. III. Drawing performed during operation on Case 2 with the duodenum rotated medially. The inset shows the close relationship of the ampulla to the proximal diverticulum.

Examination revealed a left femoral hernia which was operated upon on 8th May, 1958. At this time a cholecystogram was performed and found to be normal.

On 30th May, 1958, the patient presented with a history of "biliary colic" treated by a local doctor with injections. Examination of the abdomen revealed that she was maximally tender in the epigastrium. A barium meal on 9th June, 1958, revealed two large diverticula (Fig. IV). The proximal diverticulum appeared to arise from the outer border of the first part of the duodenum. In view of my experience with Case 2, and her history of previous stroke, I decided to be very conservative. She was given an ulcer regime to follow.

She again presented on 15th November, 1958, with further pain and I was beginning to consider operation. On 15th February, 1959, she complained that the pain was worse and that vomiting and loss of weight were worrying her. It was apparent that her condition had changed dramatically. A barium meal on 18th February, 1959, showed the two diverticula as before, but there now appeared some narrowing in the pyloric antral region suspicious of carcinoma.

Laparotomy was performed on 11th March, 1959. Multiple foci of carcinoma were found in the gut and with extensive lymph node envolvement. There was a large mass around the pyloric antral region which appeared lymphatic in origin and which

would account for the radiological appearance of narrowing. The pyloric antrum was the site of a small annular scirrhous carcinoma. There were other annular scirrhous tumours in the proximal jejunum terminal ileum, and ascending colon. The tumour of the proximal jejunum was resected for biopsy purposes. Dr. George Taylor reported that the small bowel tumour was a secondary deposit of adenocarcinoma Grade IV.



FIG. IV. The barium meal appearance of Case 3 showing the two large diverticula.

The patient remained well until 17th March, 1959, when she developed an acute abdominal pain with obvious signs of peritonitis. It was felt that the anastomosis had burst owing to obstruction caused by one of the distal gut tumours. Laparotomy confirmed these suspicions. The area of leaking anastomosis was resected again and an ileo-transverse colostomy performed. Her post-operative recovery was satisfactory but she steadily deteriorated and died two weeks later.

Autopsy confirmed the laparotomy findings and it was thought that the pyloric antral tumour was most likely to be the primary carcinoma. The duodenum was removed and the diverticula displayed (Fig. V). The very large proximal diverticulum was one of the rare true primary congenital diverticula which arise from the outer border of the first part of the duodenum.

During my autopsy dissection of the second diverticulum it was necessary to free the common bile duct closely adherent to its wall and to dissect the sac away from the pancreatic tissue in which it was embedded.

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The original dyspeptic symptoms in this patient were almost certainly due to early carcinoma and not the diverticula. attention was naturally focussed on the large diverticula, especially in view of the normal cholecystogram and otherwise normal barium meal. This case illustrates the difficulty of diagnosing gastro-intestinal carcinoma in the early stages. The autopsy specimen was unusual in that the large proximal diverticulum was of the true thick-walled variety arising from the outer border of the first part of the duodenum. Such large-mouthed diverticula are most unlikely to cause symptoms. The smaller diverticulum in the classical position arising near the ampulla illustrated the danger and difficulty liable to be met with during the dissection of such a diverticulum.



rfG. V. The autopsy specimen of Case 3 showing the unusual type of proximal diverticulum and the close relationship between the second diverticulum and the common bile duct.

In a case such as this, any attempt to excise the smaller diverticulum would have been very likely to have caused pancreatitis and maybe damage to the common bile duct which was firmly adherent to its wall. The method of spread of the carcinoma was somewhat unusual. Multiple bowel tumours were the prominent feature. It was impossible to be

dogmatic regarding the site of the primary carcinoma although the pyloric antrum would seem most likely.

#### Case 4

Diverticulum excision with cure of symptoms

Miss I.V., aged 47, was seen privately on 5th June, 1957, complaining of almost continuous nausea, upper abdominal fullness and flatulence for the past twelve months. She had suffered attacks of indigestion periodically since childhood.

Examination revealed some localized epigastric tenderness but nothing else relevant. A cholecystogram was normal. A barium meal revealed a large duodenal diverticulum which appeared to arise from the medial border of the second part of the duodenum; there was considerable stasis of barium (Figs. VI and VII).



FIG. VI. The barium meal X-ray of Case 4 showing the duodenal diverticulum.

In view of her persistent alimentary symptoms, laparotomy was performed on 6th August, 1957. A diverticulum was found arising on the medial wall of the second part of the duodenum. This was excised without difficulty. The diverticulum possessed a narrow pedicle and was of the usual thin-walled variety. There were no signs of inflammation. The patient's recovery was rapid and uneventful. Her local doctor informs me that she has been completely free of her former dyspeptic symptoms.

#### Comment

This case would seem to be the ideal and happy one in the surgical management of duodenal diverticulum. In view of the minimal pathological changes within the diverticulum it is difficult to understand how the symptoms could be explained. Time will decide whether the operation has been completely successful.

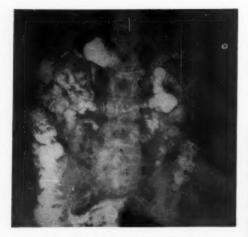


FIG. VII. The barium meal follow-through showing stasis of barium within the diverticulum.



FIG. VIII. The barium meal X-ray of Case 5 showing the diverticulum of the third part of the duodenum.

#### Case 5

Diverticulum and its most common pitfall in diagnosis

Mrs. E.M., aged 41 years, was seen in private consultation on 14th February, 1959. Her local doctor was being driven to distraction by her continued symptoms. She gave a history of epigastric pain and indigestion for several years. Occasionally the pain worried her at night. When she originally

presented to her local doctor in 1954 she seemed to have symptoms of chronic cholecystitis, although two cholecystograms were normal. She had been advised by at least two other doctors to have her gall-bladder removed. Two barium meals performed prior to 1956 were considered normal.

Her doctor reluctantly removed a normal looking gall-bladder in August, 1956. Her symptoms improved for about six months and then returned. In December, 1958, a third barium meal showed a diverticulum arising from the inner border of the third part of the duodenum (Fig. VIII). Her general practitioner endeavoured to treat her symptoms but gave up ultimately and referred her to me in February, 1959.



FIG. IX. The barium meal X-ray appearance of the gastric ulcer in Case 5. This was the fourth barium meal performed.

Examination revealed a garrulous introspective well-nourished woman. She claimed to be tender over the upper right rectus although her abdomen was difficult to examine. I could not convince myself that the small diverticulum shown on the X-ray was the explanation of her many symptoms. A fourth barium meal was arranged and on this occasion a small lesser curve gastric ulcer was found (Fig. IX).

Thus it seemed that at last we had found the true cause of her symptoms. She has been given a strict ulcer regime to follow and her local doctor tells me that she is much improved.

#### Comment

This case represents the classical instance where an accidentally discovered duodenal diverticulum has caused considerable confusion to both doctor and patient. It would have been better not to have mentioned the presence of the diverticulum to a patient so obviously introspective.

This case is also another example of a cholecystectomy performed without any good reason or benefit to the patient.

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It took three barium meals to demonstrate the diverticulum, which indicates how frequently they are missed on routine barium meal examination. Edwards (1954) found duodenal diverticula in 2 per cent. of all barium meals, but I am sure that the percenage would be higher if the barium meals were repeated on several occasions.

#### DISCUSSION

When we review the 28 cases presented, we realize how difficult it is to interpret the significance of duodenal diverticula. Fig. X summarizes the problem.

It would seem that some of the uncomplicated diverticula can cause symptoms, but it is a diagnosis to be made with extreme caution.

Waugh and Johnston (1956) showed in their 20 operative cases taken from the

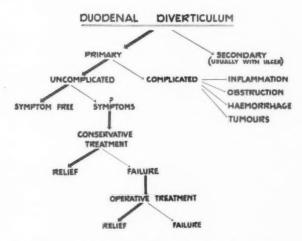


FIG. X. Chart which summarizes the problems of duodenal diverticulum.

On the fourth barium meal examination, a gastric ulcer was found after considerable difficulty. This fourth barium meal was performed by one of the most experienced and skilful of Brisbane's senior radiologists. This again illustrates the pitfalls of radiological diagnosis when carried out by the less experienced radiologist especially when hurrying to complete the volume of work given him at some of our large public hospitals.

It would seem most unlikely that the diverticulum found on X-ray examination could cause the symptoms complained of. When we look at Fig. VIII, it is apparent that such a diverticulum should empty well and would be unlikely to cause complications.

records of the Mayo Clinic between 1940 and 1952 that definite benefit from operation can be expected in little more than fifty per cent. of the most carefully selected patients. Cattell and Mudge (1952) found that an excellent result had been obtained in only 9 of 17 cases operated upon during a tenyear period at the Lahey Clinic.

In every surgical problem there are exceptions which humble us and make us reserve our dogmatism. Thus Case 1 illustrates how the complicated diverticulum can cause great confusion in diagnosis.

The uncomplicated diverticulum is so difficult to assess because the symptoms are often vague and difficult to relate to the radiological findings. Such diverticula are

usually best ignored and operation should only be considered if the diverticulum is large and retains barium after the stomach has been empty for several hours. To this we must add the likelihood that the diverticulum will cause serious complications.

Edwards (1954) advises that operation should be advised if the diverticulum is at the duodeno-jejunal flexure because in this position it is likely to cause obstructive symptoms.

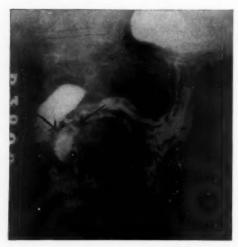


FIG. XI. A classical peri-ampullary duodenal diverticulum seen on barium meal X-ray.

Cattell and Mudge (1952) of the Lahey Clinic state that removal of duodenal diverticula is indicated in less than 5 per cent. of those discovered.

Elstner and Waugh (1957) reporting from the Mayo Clinic on 525 patients seen with duodenal diverticula state that only 8 (1.5 per cent.) underwent exploration for which the duodenal diverticulum was the primary indication.

Hilton (1954) reported a series of 9 cases in which 5 underwent diverticulectomy, 4 of whom obtained complete relief of symptoms. This illustrates how the percentage of good results will rise with increasing care in the selection of cases for operation.

When the diverticulum is complicated, there is no doubt about the necessity for operation. The chief trouble appears to be

to ascertain to what extent diverticula can cause complications. We know that certain well-known complications can arise. Case 1 illustrates how a diverticulum presumably obstructed the common bile duct. Wilbur et alii (1956) reported 6 patients in whom biliary symptoms and pathological changes appear to have been due to a peri-ampullary diverticulum. Fig. XI shows a barium meal of a classical peri-ampullary diverticulum. In one of the cases reported by Wilbur et alii (1956) a duodenal diverticulum was demonstrated by a cholangiogram after two previous operations on the biliary tract had failed to relieve pain; the patient was cured by diverticulectomy.

Thus we must admit that in patients who have had cholecystectomy with return of symptoms, or in patients who have symptoms of cholecystitis and common bile duct obstruction when no stone or stricture is found, one of the causes to be considered and looked for is a diverticulum of the duodenum.

Some of these diverticula can undergo inflammation which is a satisfactory reason for symptoms. Unfortunately many of those removed do not show evidence of inflammation or any other complication. As a result of inflammation, serious sequelae can arise. Ferguson (1953) reported a case of perforation causing peritonitis. This is a rare complication but must be considered if a preoperative diagnosis of perforated ulcer has been made and relatively little is found at laparotomy except some exudate along the anterior duodenal wall. In such a case one should explore the retro-duodenal area. Needless to say, repair in such cases may be very hazardous and difficult.

Rowlands and King (1954) reported a case of fatal abdominal haemorrhage due to erosion of a duodenal diverticulum into the adjacent aorta.

Tumours in duodenal diverticula have been reported on infrequent occasions.

One of the great problems seems to be the assessment of bleeding as a complication of duodenal diverticula.

Forrest (1957), contrary to most opinions, considered that from a review of 69 patients of duodenal diverticula, gastro-intestinal bleeding was the most common associated condition over the whole series.

In the 28 cases reviewed in this paper, 6 patients presented with either haematemesis or melaena.

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Forrest (1957) suggests that a reasonable approach to surgery for bleeding would be to consider laparotomy in patients with more than one episode of bleeding and without any other significant gastro-intestinal lesion. I feel that our experience in Brisbane suggests an even more conservative attitude to be correct.

In this paper I have avoided reference to secondary diverticula because they are usually associated with duodenal ulcers and thus the problem of management is that of the ulcer. I would like to draw attention to an instance of secondary diverticulum in the ampullary region in a patient upon whom I operated five years ago for repair of a divided common duct. As a result of the end-to-end anastomosis, and I presume, some contracture proximally, a secondary diverticulum has developed at the ampulla (Fig. XII).

# THE DIFFICULTIES AND DANGERS OF OPERATION

From my experience and that of many others, I treat the operation for excision of a duodenal diverticulum with tremendous respect. It is not a technically difficult operation if one proceeds gently. The sac is usually very thin walled and liable to tear but this does not matter because the mouth of the diverticulum is surrounded by firm tissues into which atraumatic sutures can be placed securely. The chief danger is pancreatitis and this complication can arise even though the pancreas has been minimally handled. Cattell and Mudge (1953) reported two deaths in their series of 17 operative cases due to pancreatitis.

In some cases the peri-ampullary diverticulum is embedded in the pancreas and naturally the risk of pancreatitis is very great if the sac is dissected out. In such cases I think it is better to open the duodenum and place a finger within the diverticulum to ascertain its position clearly. If the diverticulum seems embedded in the pancreas and if it possesses a wide mouth, then it is probably better to leave it alone.

The use of Banthine-like drugs and Cortisone may occasionally minimize the pancreatitis liable to ensue in cases where excision has been performed.

One of the difficulties which sometimes arise is the localization of the diverticula at opera-Carlson and Goldyne (1956) recommend the injection of air into the duodenum in those cases where the diverticulum cannot be found readily. Sometimes a duodenotomy and introduction of a finger from within helps to locate a difficult diverticulum. However in some cases, despite duodenotomy, the diverticulum may be overlooked. In one of the cases in this series the diverticulum was not found despite duodenotomy at the first operation. This was obviously due to the narrow mouth of the diverticulum making it impossible to introduce a finger from within.

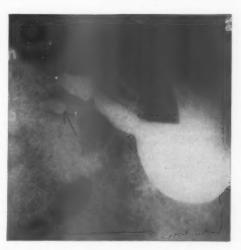


FIG. XII. A barium meal appearance of an unusual secondary duodenal diverticulum which resulted after common bile duct anastomosis and presumably traction on the ampullary region.

Various techniques for removal of these diverticula have been developed. In most cases it would seem preferable to resect the neck of the diverticulum and close the opening in two layers taking care to produce complete invagination of mucosa.

Duodenal fistula is often fatal especially if arising in the region of the ampulla.

Cattell and Mudge (1952) consider that it is advisable not to divide the diverticulum

completely flush with the duodenal wall in order to provide against leakage and to prevent narrowing of the duodenum.

When dealing with multiple diverticula, Carlson and Goldyne (1956) have shown that by-passing procedures alone are unsuccessful. However some form of by-pass such as gastro-enterostomy (with or without jejuno-jejunostomy) may be a useful adjunct to excision of multiple diverticula in an attempt to minimize pancreatitis and leakage.

#### SUMMARY

A series of 28 cases of duodenal diverticula requiring admission to hospital has been reviewed from various sources in Brisbane. Eight of these cases were subjected to excision of the diverticula with a mortality of one due to pancreatitis.

Five cases have been selected from private records to illustrate some of the problems of duodenal diverticula.

There are many pitfalls involved in both diagnosis and treatment. The incidence of bleeding in this series was surprisingly high.

It has been difficult to ascertain the effect of excision of the uncomplicated diverticulum. Many of these patients have proved very vague in their symptomatology.

Cases for operation must be selected with great care. Many barium meals may be required to establish the correct diagnosis. Cholecystectomy should be performed only if the gall-bladder is the seat of some pathological process. It was performed much too readily in this series.

Operations on duodenal diverticula are rarely justified unless complications arise. It is important to assess not only the type of diverticulum but also the type of patient in whom it is found.

#### ACKNOWLEDGEMENTS

I would like to thank Dr. A. Pye, Superintendent of the Brisbane General Hospital, and Dr. B. Purssey, Director of Medical Services at the Mater Hospital Brisbane, for permission to use hospital records. Mr. C. Leggett has allowed me to use his private records of Case 4, for which I am most grateful.

I would also like to express my thanks to Miss M. Waugh, librarian, and Miss L. Pegus, artist, of the Queensland Medical School, for their kindness and assistance. The University Photographic Department has done the photographic work entailed in this article and for their ready and efficient service I am very grateful.

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# INTRACRANIAL CHONDROMA\*

By R. F. GORMAN

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IN 1947, Forsythe, Baker, Dockerty and Camp stated that approximately 38 cases of intracranial tumours of cartilaginous structure had been reported. Few of these were pure chondromata unattached to the bones of the skull. We submit a case of solitary intracranial cartilaginous tumour, lying within the subarachnoid space, which was operated on at the Royal Perth Hospital.

Case history

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The patient, Miss S., aged 17, a stenographer, stated that four years ago she noticed the onset of transient attacks, in which the left arm and leg would become weak and slightly numb. Recently some disability of the left arm, manifested by difficulty in typing or playing the piano with her left hand, was evident between her attacks, and for the last year she had stumbled occasionally when walking. Three weeks prior to admission the frequency and severity of the attacks had increased.

#### Physical examination

Examination at the Department of Neurosurgery revealed mild upper motor neurone weakness of the left arm and leg associated with subjective loss of discrimination in the left hand. There was no evidence of raised intracranial tension, nor was any intracranial vascular murmur detected. It was considered that she had a small lesion in the parietal region of the right cerebral hemisphere, possibly a meningioma in view of the long history.

#### Special investigations

Radiography of the cranium and electro-encephalography revealed no abnormality, but right carotid angiography revealed an avascular area in the superior parietal region and slight displacement of the posterior part of the calloso-marginal artery to the left side. The avascular area was evident in both antero-posterior and lateral projections, and around this region in the late arterial phase and early venous phase of the angiogram stretched displaced vessels could be seen. These findings suggested a space-occupying lesion in the right parietal region. It was considered that these findings were due to the presence of a parasaggital meningioma.

#### Operation

The patient was operated on by Mr. J. P. Ainslie on 17th July, 1958. A parietal bone flap was reflected close to the midline. On passing the Gigli director under the bone flap, irregular projections of bone could be felt, and on reflection of the bone these

were seen to be projections of a hard tumour lying beneath the dura which was partly adherent. On reflecting the dura an irregular cartilaginous tumour lying deep to the arachnoid was revealed. tumour itself had the appearance of a chondroma, and subsequent section confirmed this diagnosis. It early became obvious that removal of the tumour intact was impracticable as irregular lobules of tumour projected deeply into the substance of the brain. The tumour cut easily with a scalpel, and a large cerebral portion of the tumour was removed by sharp dissection, after which it was possible to gently remove the many projecting lobules without serious damage to cerebral cortex. The structure of the tumour was mainly evenly cartilaginous, but the central portion was granular in appearance, suggesting calcification. A further portion had broken down to form a cystic cavity filled with oily fluid. The tumour measured 8 cms. in length and 5 cms. in width and depth. The only attachment of the tumour was by a few small vessels to the falx and dura over the longitudinal sinus.



FIG. I. Photograph of surface of tumour projecting from the brain. Reflected dura is seen in lower foreground.

Post-operatively the patient recovered consciousness quickly, but the left hemiparesis was more profound. This further increased over the ensuing twenty-four hours, but within a week had resolved and since then she has made a progressive recovery. Four months after the operation only minimal residual weakness remains.

#### Pathology of the tumour

Macroscopic: The tumour weighed 78 grams. It was composed of tough flexible semi-transparent tissue with a coarsely lobulated external surface. On the cut surfaces there were several cysts of irregular

<sup>\*</sup>Received for publication, 21st July, 1959.

outline, the largest measuring 1.3 cm. in maximum extent. The cysts contained pale or straw-coloured sticky fluid.

Microscopic: The tumour consisted of cartilage. In a few areas the cartilage cells were well preserved but in most situations they appeared to be degenerating; the cell body was swollen, the cytoplasm vacuolated, and the nucleus had lost its normal staining reaction. Cystic change was common, the cartilaginous stroma being replaced by fibrillary strands enclosing mucinous fluid. At the periphery there was a thin capsule of collagen. No meningiomatous tissue was present. The features were those of a chondroma.



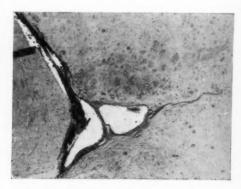
FIG. II. Tumour showing classical cartilage appearance.

#### DISCUSSION

Intracranial chondromata are seldom encountered surgically, and it is unlikely that a correct diagnosis will be made pre-operatively, but if one considers the possibility of such a tumour diagnosis may be suggested by the extreme avascularity of the lesion as seen on angiography, there being a complete absence of abnormal vessels as occurs in meningiomata, and the pattern of normal vessels seen suggested irregular lobulation also unusual in meningiomata.

The lobulation of the tumour makes removal difficult, and one must resist the temptation to remove an interesting pathological specimen intact.

Tumours which show histological differentiation into cartilage have been seen as primary tumours in various parts of the intracranial cavity and have been reported in the position of the case reported, with a similar attachment to the dura or to the falx. Chondromata have been described arising from the bones of the base of the skull, in the tela choroidea of the ventricles, and in the posterior fossa. Letterer (1920) describes two cases of chondromata of the ventricles, and Freiman and Ficarra (1943) report on osteochondro-blastic meningioma of the left cerebellar hemisphere. Elsberg (1926) reports a case of an encapsulated chondroma pressing on the trigeminal ganglion, but does not state whether it was attached to the base of the skull.



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FIG. III. Lowpower photomicrograph: The surface of the tumour is separated from brain (left) by its capsule and thin-walled blood vessels.

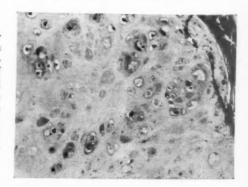


FIG. IV. Highpower photomicrograph showing cartilaginous structure.

# Pathology

The above-mentioned tumours are of varying histological structure and graded malignancy, but if we subscribe to the theory of Alpers (1935), wherein it is believed that these tumours originate from metaplasia of

the arachnoid and choroid plexus, then the aggregation of these tumours together is a fair one. Alpers states that unless the dural attachment is removed, they tend to recur. The dural attachment in this patient was so insignificant that recurrence appears most improbable.

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With regard to the theories of origin, it is convenient to consider these tumours in the three main sites in which they are found, namely, the base of the skull, choroid plexus and meninges. It is not surprising that the base of the skull should give rise to intracranial chondromata because the occipital temporal, sphenoid and ethmoid bones, together with the inferior concha, are developed wholly or partly from cartilage. As far as the other two sites are concerned, the reason for the occurrence of cartilaginous tumours is not quite so obvious. It is recognized that in the adult organism there is persistance of fibroblastic cells with the potencies of undifferentiated mesenchymal cells, and these may undergo progressive development and furnish new cell types (Maximow and Bloom, 1942). These pluripotential fibroblasts would thus be capable of producing chondrocytes. An explanation for the chondroma attached to the dura is furnished by the fact that the outer layer of the dura forms an internal periosteum for the bones of the skull and all periosteum has the ability to form cartilage (Chorobski et alii, 1939).

#### SUMMARY

- Solitary intracranial chondroma is a very rare condition.
- Herein we have reported this condition in a 17-year-old female.
- Theories of pathogenesis are briefly discussed.

#### ACKNOWLEDGEMENTS

I wish to thank Mr. J. P. Ainslie for his invaluable advice and help in the preparation of this article. I am indebted to Dr. R. Findlay Jones, Pathologist to the Royal Perth Hospital, for his competent appreciation of the pathological aspects. The members of the staff of the Neurosurgical Unit, Photographic and Pathology Departments of the Royal Perth Hospital were most co-operative and materially assisted in presenting this case.

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# A FATAL CASE OF CANCER OF THE GALL-BLADDER† SOME AETIOLOGICAL AND SURGICAL OBSERVATIONS

By T. S. REEVE\* AND F. F. RUNDLE

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CANCER of the gall-bladder is an almost universally fatal disease; Pack et alii (1955) reported that, in the entire history of the Memorial Cancer Center, there had been only one five-year survival after surgery. Here we summarize certain aetiological and therapeutic aspects of this cancer as they were illustrated in a noteworthy patient, observed and treated in this hospital over a period of thirty-four years.

The patient, a married woman with 3 children, was admitted on six occasions as follows:—

(1) In 1925, for cholecystostomy and removal of stones

She was then aged 32 and had already suffered from indigestion and flatulence for years. Numerous calculi were removed from the gall-bladder. After the operation, she entered on a prolonged period of good health, marred only by mild flatulent dyspepsia.

(2) In August, 1953 (twenty-eight years later), for drainage of an abscess in the abdominal wall

Four months previously she developed right upper abdominal pain which had become progressively more severe and constant. A large tender swelling lay deep to the old incision and the skin overlying it was reddened. No gall-stones, but a large amount of pus, growing E. coli, was evacuated. Subsequently a fistula formed in the scar. X-rays showed the gall-bladder to be non-functioning.

(3) In October, 1953 (three months later), for cure of fistula and cholecystectomy (carcinoma of gallbladder)

The fistulous track in the skin and subcutaneous tissues was excised down to the scar in the aponeurosis and muscle but was not followed more deeply because of its irregular course. The gall-bladder was enlarged and thick-walled. It was removed without difficulty and not until it was laid open afterwards was carcinoma diagnosed (Fig. I).

(4) In February, 1957 (three and one-third years later), for excision of a recurrence in the scar)

At a routine follow-up she stated that she had been quite well, but on examination a rounded lump

about 3" in diameter was discovered lying in the plane of the muscles of the abdominal wall. It was excised widely leaving a defect, approximately 8" x 6", which was closed with a large tantalum gauze prosthesis. Examination of the liver, pancreas, and other abdominal contents at operation showed no evidence of other recurrence or metastasis.



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FIG. I. The operative specimen laid open to demonstrate the tumour and its extensive mucosal infiltration.

(5) One year afterwards (February, 1958), for biopsy of mass in right tibia

She had complained of swelling and pain in the right shin for four months and X-rays showed "sunray" periostitis in its middle-third with some destruction of cortical bone (Fig. II). A biopsy showed "columnar cell carcinoma consistent with a primary lesion in the gall-bladder" (Fig. III). The area

<sup>\*</sup>Senior Surgical Research Officer, working with the aid of a grant from the New South Wales State Cancer Council.

<sup>†</sup>Received for publication, 20th July, 1959.

was irradiated. Metastasis to bone is evidently unusual; it is not mentioned in any of the 72 cases recorded by Burdette (1957).

(6) After six months (September, 1958), in a state of terminal carcinomatosis

At autopsy the pancreas was found to be diffusely infiltrated by carcinoma, multiple large masses of metastatic cancer were present in the liver; among the bones the right tibia and skull showed extensive involvement by well-differentiated columnar cell carcinoma.

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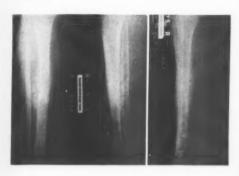
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FIG. II. (a) X-ray of the right tibia, 2nd May, 1958, demonstrating metastatic tumour following a course of irradiation.

(b) X-ray of the right tibia taken several weeks before death, further extension of the tumour and the development of sun-ray appearance are evident.

# COMMENT

There is an undeniable association between cholelithiasis and carcinoma of the gallbladder (Burdette, 1957). Thus, in different series, stones are reported to have been present in 60-100 per cent. of cases (Mohardt, 1939; Donhauser, 1958). In many specimens stones are not specifically mentioned or looked for; thus the true figure may be nearer 100, than 60 per cent. Approached from the other aspect, calculous gall-bladders have been found also to be the site of carcinoma in 1 per cent. of patients submitted to biliary surgery (Kirshbaum and Kozoll, 1941; Ulin et alii, 1950). This figure may overstate the actual association since many calculous gallbladders are never removed but few containing tumours remain undiscovered.

It is likely that, in the present patient, empyema of the gall-bladder and an abdominal wall abscess in the old cholecystostomy tract, resulted from blockage of the cystic duct by growth. The extent of involvement of the mucosal aspect is indeed noteworthy, in view

of the lack of invasion through the gall-bladder wall at any point. Thus the diagnosis was not made until after routine cholecystectomy. It is significant also that recurrence did not take place in the gall-bladder fissure or its neighbourhood.

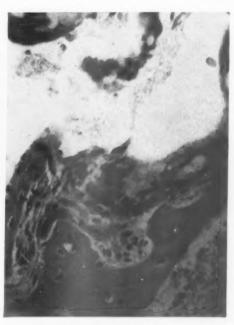


FIG. III. Photomicrograph of tissue from the tibia; adenocarcinoma is seen invading the bone marrow between islands of bone. (x 400)

The mucosal cancer may also have spread into the deepest part of the fistulous track which, unfortunately, was not excised. Alternatively, loose fragments of growth may have lodged and grown there, ultimately producing a large spherical recurrence in the abdominal wall. Empyema and perforation are well recognized complications of gall-bladder cancer, and a sinus track was present in 3.6 per cent. of Donhauser's cases (1958). Carcinoma extended along the old sinus track forming after cholecystostomy, in two cases of carcinoma reported by Finney and Johnson (1945).

In addition to the need for complete excision of all such fistulous tracks, this patient's long history and its fatal outcome re-emphasize certain surgical lessons. Removal of stones by cholecystostomy does not protect the patient from gall-bladder cancer, even when no further stones form. In one of Willis' (1942) patients, cancer developed twenty-four years after the removal of gall-stones by cholecystostomy. In the present patient the interval was twenty-eight years. Among a series of 45 cases of gall-bladder cancer reported by Roberts (1954) no fewer than 7 had previously undergone cholecystostomy; in one the operation was done forty-five years previously. No mention is made however of whether or not stones had reformed in that case.

Is routine cholecystectomy for stones justifiable prophylaxis against cancer of the gall-bladder? This is hardly a problem where the stones are symptomless. And, clearly, where they are producing symptoms, cholecystectomy is a safe routine not so much as a prophylaxis against cancer, the risk of which is slight, as against other more frequent complications of the stones themselves.

#### SUMMARY

A patient who underwent cholecystostomy and removal of multiple gall-stones, developed, twenty-eight years later, an empyema of the gall-bladder, an abdominal wall abscess and fistulous track. The underlying condition was a cancer in the gall-bladder. Noteworthy features were the long latent period of twenty-eight years between cholecystostomy and her presenting with cancer, the subse-

quent development of a massive recurrence in the abdominal wall three and one-third years after cholecystectomy, and the probability that this recurrence resulted from extension of the growth to the fistulous track. Failure to excise the track completely may have determined the eventually fatal outcome five years after cholecystectomy.

#### ACKNOWLEDGEMENTS

Our thanks are due to the General Medical Superintendent of the Royal North Shore Hospital for permission to report this case. er gl st cl op P m F in

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The photographs were taken by Miss M. Simpson, Clinical Photographer, Royal North Shore Hospital of Sydney.

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# Books Reviewed

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By PETER F. HALL. Edition: Monographs of Federal Council of the British Medical Association, No. 2. Sydney, Australia: Australasian Medical Publishing Co. Ltd., 1959. 10" x 7½", 157 pp., 24 figures. Price: £1 15s.

The best definition of gynaecomastia should be expressed in simple terms: "enlargement of the glandular component of the breast in males," for such a definition is free of contention to both clinicians and surgeons. This is the considered opinion of Dr. Peter F. Hall, Honorary Assistant Physician to the Sydney Hospital, in his splendid monograph "Gynaecomastia," a publication by the Federal Council of the British Medical Association in Australia.

In sixteen excellent chapters Dr. Hall reviews the early descriptions of the malady and then describes its association with physiological states, diseases of the endocrine glands, cirrhosis of the liver, malnutrition, the taking of certain drugs, diseases of the nervous system and lungs, and genetic factors.

In the chapter on diseases of the liver, Dr. Hall describes the part played by the normal liver in destroying oestrogens, and its failhre to do this in cirrhosis. But he fails here to consider the part played by malnutrition per se in the production of both gynaecomastia and cirrhosis. Later, however, he describes the association of gynaecomastia with malnutrition and refers to the excellent studies of Kark, Morey and Paynter (1951) which revealed that gynaecomastia not only occurred in cirrhosis of the liver caused by prolonged malnutrition, but was most conspicuous immediately after the period of malnutrition, when there was a return to normal diet. This produced a return of full sexual activity and often gynaecomastia.

Klinefelter's syndrome, characterized by small testes, ozospermia, raised levels of gonadotrophins and gynaecomastia, is reviewed in the light of chromosomal sexing and it is submitted that distinct clinical and genetic groups can now be identified. It is of importance for readers to recall that recently Klinefelter's syndrome has been thrown into prominence by the demonstration by Jacobs and Strong (1959) that the males suffering from this disease have 47 chromosomes in the nuclei in place of the normal 46.

And finally this monograph contains a wise chapter on treatment of gynaccomastia. The clinician is urged to seek the cause in each individual case and then endeavour to remove or modify it. But in deciding the optimum treatment the clinician must be confident that the patient will be made happier by the "cure." Thus liberal surgical removal of breast tissue may cause the patient great displeasure.

This book is lucid and the illustrations liberal and well produced. It is commended to all physicians practising general medicine, and especially to those who have made a special study of the endocrine glands and psychiatry. Moreover surgeons will be well advised to read the monograph before performing the Jerome Webster operation of partial mastectomy, an operation first practised by Paulus Ægineta in the seventh century A.D.

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JACOBS, P. A. and STRONG, J. A. (1959), Nature (Lond.), vol. 183, page 302.
KARK, R. M., MOREY, G. R. and PAYNTER, C. R. (1951), Amer. J. med. Sci., vol. 222, page 154.

PROCEEDINGS OF INTERNATIONAL CONFERENCE ON THE INSULIN TREATMENT OF PSYCHIATRY, held at New York Academy of Medicine, 24th-25th October, 1958

Edited by Dr. MAX RINKEL, M.D., and Dr. HAROLD HIMWICH, M.D. New York, U.S.A.: Philosophical Library, New York, 1959. xxix plus 386 pp., 8½" x 5½". Price: \$5.00.

Almost a year after the death of Manfred Sakel, the discoverer of hypoglycaemic insulin treatment of schizophrenia, the Proceedings of an International Conference on Insulin Treatment in Psychiatry, held in New York in 1958, has been published.

"Proceedings of International Conference on the Insulin Treatment in Psychiatry held at New York Academy of Medicine" was edited by Drs. Max Rinkel and Harold Himwich. It is a collection of papers presented at the Conference. The contributors are world authorities on insulin treatment, representing the best views from Austria, U.S.A., Argentina, Peru, Brazil and Great Britain. This is not a textbook; it is a recorded account of both the main papers and the discussions in extenso.

The purpose of the Conference was to assess the value of insulin in the treatment of psychoses. is timely, as there is little doubt that the popularity of this form of treatment is on the wane, certainly in this country, and in the United States of America and Great Britain. The reasons for this are discussed and it is submitted that the chief one is that the failures in treatment leading to this unpopularity result from variations of the therapeutic ritual of Sakel. Insulin therapists who read these proceedings may not agree that these variations, which certainly do occur, would in fact account for the difference in results of insulin therapy. Not all would share the persisting enthusiasm of the Viennese School. All aspects of treatment are covered and include its history, psychology, endocrinological and biochemical aspects and indications and results. present trends, together with the personal experiences in various centres are compared. Attempts are made to explain the theory of insulin therapy, in particular by O. H. Arnold. The schizophrenic processes are regarded by him as a heredo-genetically preformed bottleneck leading to dysfunction of single groups of ganglion cells which are responsible for the control of thalamo-commissural processes.

The "bottleneck" is said to reside in either the anoxybiotic phase of carbohydrate metabolism or in relation to that aspect of metabolism related to the balance of "acceptors and donators of phosphates." Arnold assumes that affected single cells or units must be destroyed. He believes that insulin causes disorganisation within these ganglion cells. He suggests that both E.E.G. and post-mortem conclusions support this thesis. The "moulting" effect produced by such destruction presumably allows normal cells to operate at an optimum level.

The summary of the observed biochemical changes, by Himwich, makes much more impressive reading, William Sargeant's contribution is concerned with the relationship of insulin to other therapies, and is a workmanlike assessment of the position by a psychiatrist who obviously appreciates the strengths and weaknesses of all physical therapy. He finds insulin best combined with electroshock given in the early stagts of coma. He is impressed with the good effects resulting from the induction of both severe brain excitement and deep coma and collapse. He regards this excitement as a helpful and abreactive agent. It is clear that Sargeant is prepared to use any of the current physical methods of treatment for schizophrenia and by no means accepts insulin as the answer.

Some carefully prepared figures and results of insulin treatment are presented which suggest that remission to some degree may be expected in up to 70 per cent. of cases, with a higher rate of "total remission." 52 per cent. in catatonic types, ranging to a (surprisingly high) figure of remission of 10 per cent. for simple primary types.

In general these proceedings will be mainly of interest to psychiatrists who use insulin as a therapeutic agent. It is doubtful if they will convert the non-believer. Its presentation is of necessity too disjointed for the novice. Scant attention is paid to the advantage of psychotherapy. More severe editing of the discussion would have shortened the text with advantage.

This book is a fitting tribute to the pioneering work of Manfred Sakel.

#### THE LIFE AND TIMES OF SIR CHARLES HASTINGS.

By WILLIAM H. McMENEMEY, M.A., D.M., F.R.C.P., D.P.M. Edinburgh and London: E. & S. Livingstone Ltd., 1959. 8\frac{1}{2}" x 6\frac{1}{2}", xii plus 516 pp., frontispiece and 32 plates. Price: 50s. (stg.).

In 1951 Dr. McMenemey delivered the Hastings Memorial Oration. The material which he collected has now been elaborated into a book. Charles Hastings (1794-1866), a Worcester practitioner, was instrumental in the foundation of the British Medical Association which he commenced in the provinces as the Provincial Medical and Surgical Association in 1832 and which twenty-four years later, was to change its name to the one so familiar today. He also worked hard for reform of the medical profession in general, his work gaining official recognition by the passing of the Medical Act of 1858. This Act enabled the formation of the General Council of Medical Education and Registration on which Hastings served from its inception until 1863.

Dr. McMenemey has collected a vast amount of detailed information on Hastings' life, his contemporaries and medical practice of the period. This makes most interesting reading even although, on occasions, the thread of his story may be partly hidden by the very weight of his material. Nevertheless this volume will certainly be, for all time,

the definitive biography of a man who spent his whole life seeking to improve medical practice and the lot of his professional colleagues. In this book it is interesting to note the influence and power wielded by Thomas Wakely, of the Lancet, sometimes critical of Hastings, sometimes supporting his efforts. The volume is very well produced and profusely illustrated.

#### INTESTINAL OBSTRUCTION.

By CLAUDE E. WELCH, M.D., D.Sci.(Hon.). Chicago, U.S.A.: The Year Book Publishers Inc., 1958. 9" x 6", 376 pp., 135 illustrations. Price: \$10.50.

This monograph presents a good review of the problems of obstruction of both the large and small intestines. The style is sometimes a little irritating, and some of the statements are ingenuous; but the book is comprehensive and contains some fresh ideas.

In the clinical picture of small bowel obstruction, Dr. Welch rightly stresses the value of the stethoscope—"auscultation furnishes the most important information." However, few would agree that the interval between waves of colicky pain is always "2 to 3 minutes."

Most surgeons will be glad to see that he advocates open anastomosis when resection is necessary, but few will admire the rather traumatic clamps with which he holds the bowel ends.

In the operative management of large bowel obstruction, Dr. Welch provides a full and detailed account of "obstructive resection," but surely this is an obsolete operation.

In the section on replacement therapy, we are surprised to learn that "most surgical patients will not require any sodium in the post-operative course."

There is a regrettable tendency to enlarge on operative detail in such procedures as excision of the rectum, in a fashion not relevant to the theme of the book.

It is interesting to find that a thoraco-lumbar sympathectomy is advocated for recurrent small bowel obstruction.

On the whole, this is an interesting book for those who are concerned with the theoretical aspects of this problem.

### A MANUAL OF ANAESTHETIC TECHNIQUES.

By WILLIAM J. PRYOR. Second Edition. Bristol, England: John Wright & Sons Ltd., 1959.  $8\frac{1}{2}$ "  $\times$   $5\frac{1}{2}$ ", 228 pp., 75 illustrations. Price: 27s. 6d. (stg.).

The fact that a second edition of this book has appeared in three years indicates that a demand exists for it. Though much of the original material is retained, some changes have been made in order to bring the text up to date. The references are now in a more useful form and enable the reader to refer to original papers.

This book should continue to find favour with resident anaesthetists and registrars as it gives, in brief, standard methods of coping with most situation.

# Book Received

# MORE MEDICAL AND OTHER VERSES.

By ALEX. E. ROCHE. First Edition. London: H. K. Lewis & Co. Ltd., London. 7½" x 4¾", viii plus 35 pp. Price: 5s. (stg.).

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